THE UNIVERSITY OF QUEENSLAND

AUSTRALIA

Oropharyngeal dysphagia in preschool children with cerebral palsy: relationship to gross motor function, dietary intake, and nutritional status

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Master of Public Health, Bachelor of Speech Pathology

A thesis submitted for the degree of Doctor of Philosophy at

The University of Queensland in 2015

School of Medicine

Abstract

Context: Oropharyngeal dysphagia (OPD) is common in preschool children with cerebral palsy (CP), and may negatively influence children's dietary intake and nutritional status. Prevalence estimates range from 19% to 99%, with this large variability owing to study methodology. Most studies detected OPD through parent report, and recruitment has focused on children with moderate-severe CP and from a broad age range. Understanding the prevalence and patterns of OPD in preschool children with CP across the full range of gross motor functional levels will promote earlier detection and interventions.

Objective: The broad aim of this doctoral research was to determine the prevalence and patterns of OPD in preschool children with CP from 18 to 36 months; and its relationship to dietary intake, nutritional status and gross motor function.

Design: This doctoral research forms part of 2 larger longitudinal cohort studies, CP Child: Growth, Nutrition and Physical Activity (GNPA); and CP Child: Brain Structure and Motor Function. Four substudies comprise this doctoral thesis: (1) systematic review of OPD measures, and validity and reproducibility, (2) cross-sectional studies of OPD, (3) longitudinal study of OPD, (4) cross-sectional study of OPD in a low-resource country.

Participants: Participants in all substudies were children with a confirmed diagnosis of CP aged 18 to 36 months corrected age. One hundred and thirty children participated in the main GNPA sample; inclusion of Queensland-born children from birth years 2006-2009, and exclusion of children with neurodegenerative conditions or syndromes influencing growth. Forty children with typical development (TD) were recruited as a reference sample. Eighty-one Bangladesh-born children were recruited to the sample from a low-resource country.

Procedure: Children attended the hospital for mealtime and gross motor assessment, and growth anthropometry. Mealtimes were evaluated using the *Schedule for Oral Motor Assessment* (SOMA), *Dysphagia Disorders Survey* (DDS), *Pre Speech Assessment Scale* (PSAS), 16 clinical signs suggestive of pharyngeal phase impairment, and the *Thomas-Stonell & Greenberg Saliva Severity Scale*. Parents reported on their child's mealtime using the *Queensland CP Child Feeding Questionnaire*, which was developed for the study. Gross motor function was classified on the *Gross Motor Function Classification System* (GMFCS), motor type (spasticity, dyskinesia, ataxia and hypotonia) and distribution. Parents completed a 3-day weighed food record at home, from which

dietary intake was calculated. Nutritional status was indicated by height, weight, and body mass index, converted to z scores using age and gender reference data.

Results: A systematic review of the clinimetric properties of OPD measures identified the SOMA and DDS to have the strongest psychometric properties and clinical utility. Our validity and reproducibility substudy found the SOMA, DDS and PSAS to all have strong reproducibility (agreement >85%, κ >0.5). The SOMA had the best specificity (100%), but reduced sensitivity (53%); whereas the DDS and PSAS had high sensitivity (100%) but reduced specificity (47% and 71%, respectively). Modified OPD cut-points were calculated for each measure based on a high prevalence of OPD in children with TD.

OPD prevalence based on 1 or more measures (SOMA, DDS, clinical signs) was identified in 85% of preschool children with CP. The prevalence estimate calculated using latent-class methods was 65%, and estimates using the modified cut-points ranged from 46% (PSAS) to 62% (SOMA). OPD was prevalent across all levels of gross motor function, with a stepwise increase in the proportion with OPD with increasing GMFCS level. Children who were nonambulant (GMFCS V) had significantly increased odds of OPD compared to those who were ambulant (GMFCS I) (OR = 17.9, *P* = .036). Almost all children had oral phase impairments (94%, using modified cut-points 79%). The proportion of children with clinical signs suggestive of pharyngeal phase impairments was lower (68%, using modified cut-points 51%).

Longitudinally, the prevalence of OPD reduced marginally between 18 to 24 months and 36 months, from 62% to 59% (n=53). The greatest number of children whose OPD improved were from GMFCS I (n=6, 27%), although the greatest proportion of a GMFCS level were children from GMFCS IV (n=3, 75%). GMFCS was the only risk factor which was consistently associated with OPD at both assessment points.

OPD prevalence (based on DDS modified cut-points) was greater in Bangladesh (total n=81, 68%) compared to Australia (total n=130, 56%). However, prevalence and severity did not differ significantly between high- and low-resource countries when stratified for GMFCS (prevalence OR=2.4, *P* = .051; severity β=1.2, *P* = .08).

Conclusions: The findings support that OPD is prevalent in about 60% of preschool children with CP, and is present even in children with ambulatory CP (GMFCS I-II). GMFCS was the strongest predictor of OPD in preschool children with CP, and this persisted across time, and in different resource and ethnic contexts. This thesis provides useful information as a basis for earlier identification of children at risk of growth or respiratory consequences associated with OPD, as well as to assist in planning optimal oropharyngeal sensorimotor therapies and nutritional interventions.

Declaration by author

This thesis is composed of my original work, and contains no material previously published or written by another person except where due reference has been made in the text. I have clearly stated the contribution by others to jointly-authored works that I have included in my thesis.

I have clearly stated the contribution of others to my thesis as a whole, including statistical assistance, survey design, data analysis, significant technical procedures, professional editorial advice, and any other original research work used or reported in my thesis. The content of my thesis is the result of work I have carried out since the commencement of my research higher degree candidature and does not include a substantial part of work that has been submitted to qualify for the award of any other degree or diploma in any university or other tertiary institution. I have clearly stated which parts of my thesis, if any, have been submitted to qualify for another award.

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Publications during candidature

Peer Review Papers

1. **Benfer KA**, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Food and fluid texture consumption in a population-based cohort of preschool children with cerebral palsy: relationship to dietary intake. *Developmental Medicine and Child Neurology* 2015; http://dx.doi.org/10.1111/dmcn.12796. Accessed May 15, 2015.
2. **Benfer KA**, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Clinical signs suggestive of pharyngeal dysphagia in preschool children with cerebral palsy. *Research in Developmental Disabilities* 2015; 38:192-201.
3. **Benfer KA**, Jordan R, Bandaranayake S, Finn C, Ware RS, Boyd RN. Motor severity in children with cerebral palsy studied in a high-resource and low-resource country. *Pediatrics* 2014; http://pediatrics.aappublications.org/content/early/2014/11/18/peds.2014-1926.abstract. Accessed November 24, 2014.
4. **Benfer KA**, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Validity and Reproducibility of Measures of Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy. *Developmental Medicine and Child Neurology* 2014; http://dx.doi.org/10.1111/dmcn.12616. Accessed November 16, 2014.
5. **Benfer KA**, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Oropharyngeal dysphagia in preschool children with cerebral palsy: oral phase impairments*. Research in Developmental Disabilities* 2014;35:3469-3481.
6. **Benfer KA**, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Oropharyngeal Dysphagia and Gross Motor Skills in Children with Cerebral Palsy. *Pediatrics* 2013:e1553-e1562.
7. **Benfer KA**, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Longitudinal cohort protocol study of oropharyngeal dysphagia: relationships to gross motor attainment, growth and nutritional status in preschool children with cerebral palsy. *BMJ Open* 2012;2(4):e001460. http://bmjopen.bmj.com/content/2/4/e001460.full.pdf. Accessed August 27, 2012.
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9. **Benfer KA**, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Longitudinal study of oropharyngeal dysphagia in preschool children with cerebral palsy. Under review with Research in Developmental Disabilities.

10. **Benfer KA**, Weir KA, Bell KL, Davies PSW, Ware RS, Boyd RN. Oropharyngeal dysphagia in children with cerebral palsy studied in a high and low resource country. Under review with Developmental Medicine and Child Neurology.

Peer Review Abstracts

1. **Benfer KA**, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy: Comparison between High- and Low-Resource Countries. *Developmental Medicine and Child Neurology* 2014;56(Supp 5):78-79. (Abstract)
2. **Benfer KA**, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Dietary Intake and Undernutrition in Preschool Children with Cerebral Palsy: Comparison between High- and Low-Resource Countries. *Developmental Medicine and Child Neurology* 2014;56(Supp 5):5-6. (Abstract)
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preschool aged children with cerebral palsy. *Developmental Medicine and Child Neurology* 2013;56(Supp 2):3-4. (Abstract)

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Contributor	**Statement of contribution**
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Benfer KA, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Longitudinal cohort protocol study of oropharyngeal dysphagia: relationships to gross motor attainment, growth and nutritional status in preschool children with cerebral palsy. *BMJ Open* 2012;2(4):e001460. http://bmjopen.bmj.com/content/2/4/e001460.full.pdf. Accessed August 27, 2012. – Incorporated as Chapter 3

Contributor	**Statement of contribution**
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Benfer KA, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Oropharyngeal Dysphagia and Gross Motor Skills in Children with Cerebral Palsy. *Pediatrics* 2013:e1553-e1562. – Incorporated as Chapter 4.

Contributor	**Statement of contribution**
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Davies, PSW	Editorial guidance (10%) Doctoral supervision (10%)
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Benfer KA, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Validity and Reproducibility of Measures of Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy. *Developmental Medicine and Child Neurology* 2014; http://dx.doi.org/10.1111/dmcn.12616. Accessed November 16, 2014. – Incorporated as Chapter 5

Contributor	**Statement of contribution**
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Benfer KA, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Food and fluid texture consumption in a population-based cohort of preschool children with cerebral palsy: relationship to dietary intake. *Developmental Medicine and Child Neurology* 2015; http://dx.doi.org/10.1111/dmcn.12796. Accessed May 15, 2015 – Incorporated as Chapter 6

Contributor	**Statement of contribution**
Benfer, KA (Candidate)	Data extraction/ rating (feeding videos) (100%) Analysis and interpretation of data (80%) Manuscript writing (100%)
Weir, KA	Editorial guidance (35%) Doctoral supervision (30%)
Bell, KL	Editorial guidance (10%)
Ware, RS	Statistical advice (20%) Editorial guidance (10%)
Davies, PSW	Editorial guidance (10%) Doctoral supervision (10%)
Boyd, RN	Editorial guidance (35%) Doctoral supervision (60%)

Benfer KA, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Oropharyngeal dysphagia in preschool children with cerebral palsy: oral phase impairments*. Research in Developmental Disabilities* 2014;35:3469-3481 – Incorporated as Chapter 7

Contributor	**Statement of contribution**
Benfer, KA (Candidate)	Data extraction/ rating (feeding videos) (100%) Analysis and interpretation of data (80%) Manuscript writing (100%)
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Davies, PSW	Editorial guidance (10%) Doctoral supervision (10%)
Boyd, RN	Editorial guidance (35%) Doctoral supervision (60%)

Benfer KA, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Clinical signs suggestive of pharyngeal dysphagia in preschool children with cerebral palsy. *Research in Developmental Disabilities* 2015; 38:192-201 – Incorporated as Chapter 8

Contributor	**Statement of contribution**
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Benfer KA, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Longitudinal study of oropharyngeal dysphagia in preschool children with cerebral palsy. Under review with Research in Developmental Disabilities – Incorporated as Chapter 9

Contributor	**Statement of contribution**
Benfer, KA (Candidate)	Data extraction/ rating (feeding videos) (100%) Analysis and interpretation of data (80%) Manuscript writing (100%)
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Boyd, RN	Editorial guidance (35%) Doctoral supervision (60%)

Benfer KA, Jordan R, Bandaranayake S, Finn C, Ware RS, Boyd RN. Motor severity in children with cerebral palsy studied in a high-resource and low-resource country. *Pediatrics* 2014; http://pediatrics.aappublications.org/content/early/2014/11/18/peds.2014-1926.abstract. Accessed November 24, 2014. – Incorporated as Chapter 10

Contributor	**Statement of contribution**
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Benfer KA, Weir KA, Bell KL, Davies PSW, Ware RS, Boyd RN. Oropharyngeal dysphagia in children with cerebral palsy studied in a high and low resource country. Under review with Developmental Medicine and Child Neurology – Incorporated as Chapter 10

Contributor	**Statement of contribution**
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Davies, PSW	Editorial guidance (10%) Doctoral supervision (10%)
Boyd, RN	Editorial guidance (35%) Doctoral supervision (60%)

Contributions by others to the thesis

Conception, design, ethics and grant applications for Growth Nutrition and Physical Activity project: Peter Davies, Roslyn Boyd, Kristie Bell, Kelly Weir, Richard Stevenson
Design of feeding questionnaire: Kelly Weir
Recruitment and collection of feeding videos: Laura Pareezer, Rachel Jordan, Christine Finn, Lauren Forbes, Joanne McMah, Jacqueline Walker, Stina Oftedal
Nutritional analysis of food records: Lauren Forbes, Joanne McMah, Jacqueline Walker, Stina Oftedal
Classification of gross motor function: Rachel Jordan, Christine Finn, Roslyn Boyd
Inter-rater ratings (for systematic review and oropharyngeal dysphagia measures): Kelly Weir

Statement of parts of the thesis submitted to qualify for the award of another degree

The work completed for this doctoral thesis builds upon a thesis submitted for the award of a Masters of Public Health (MPH), at LaTrobe University (awarded in 2011). No publications arose from the MPH thesis. A major contribution added by this thesis was the inclusion of a longitudinal substudy, whereby children were reassessed 12 to 18 months after their initial assessment. Also, in order to delineate some of the limitations to ingestion functions associated with typical development, a reference sample was included. Furthermore, the sample size of the main study in this PhD thesis is almost double that which was included in the MPH thesis, and covers a broader age range. These additions have allowed significantly more in depth analyses of the topic, including detailed testing of the validity of measures, which has assisted in understanding the construct of OPD. This will allow greater generalisability of the thesis findings, and enable better interpretation and clinical application of the results.

Acknowledgements

I would like to thank my advisory team who have guided and supported me throughout this doctoral research; and for the many hours they have invested in my learning, and in improving the quality of my work.

Prof Roslyn Boyd, my primary advisor, for her commitment to all aspects of this project, for her vision and innovation, prioritisation of my learning in her busy schedule, and support with the dissemination of the research findings (including publications and attendance at conferences).

Ms Kelly Weir, for her commitment and encouragement, for the discussions and her advice, her specialised knowledge in paediatric dysphagia, and her carefully considered feedback.

Prof Peter Davies, for his vision as Chief Investigator of the Growth, Nutrition and Physical Activity study; for his support particularly surrounding the nutritional aspects of this project; and for his questioning and sparking of broader research enquiry.

I would also like to thank my colleagues, past and present, at the Queensland Cerebral Palsy and Rehabilitation Research Centre, The University of Queensland:

Dr Kristie Bell, study coordinator and co-author, for her overall study guidance and contribution to the nutritional aspects of the study.

Dr Robert Ware, for his statistical guidance, and particularly for his quick responses to emergency emails.

Ms Laura Pareezer, Ms Rachel Jordan, Ms Christine Finn, Ms Lauren Forbes, Ms Joanne McMah, Ms Stina Oftedal, Dr Jacqueline Walker, Ms Camilla Davenport, Dr Andrea Coleman, Ms Naomi Westwood, and the administration team; for their significant support surrounding logistics, recruitment, dietary and gross motor ratings, and data retrieval.

Thank you to my colleagues and supporters in Bangladesh, who provided invaluable support enabling the Bangladesh substudy.

Staff at the Centre for the Rehabilitation of the Paralysed; I would like to thank Dr Valerie Taylor, Md. Shafiq-ul Islam, Jannatul Ferdous, Ms Sharmin Hasnat, Md. Jahangir Alam, Md. Morshedul Quadir, Dr Sabera Bilkis, Ms Hosneara Parveen, Ms Shoma.

Dr Baitun Nahar and Dr Tahmeed Ahmed at the International Centre for Diarrhoeal Disease Research.

Dr Naila Khan and her team at the Dhaka Shishu Hospital.

I would like to acknowledge the financial support provided to me and my projects by the National Health and Medical Research Council, Speech Pathology Australia, and the Graduate School of the University of Queensland.

Particular thanks to all those who have loved, encouraged, guided and nudged me along my journey to reach this point. My partner Andrew, family, friends, community, work colleagues, and academics who have all fostered my curiosity of spirit and love of learning, and have supported me throughout this process.

Finally, a special thank you to all the children and their families, both in Australia and Bangladesh, who have made time to participate, and endured the hours of travel, countless questionnaires and weighed food records; all of which has made this research possible. I hope that your efforts will contribute to better nutritional and health outcomes for children with cerebral palsy in the future.

Keywords

oropharyngeal dysphagia, deglutition disorders, cerebral palsy, dietary intake, gross motor, paediatrics, validity and reliability, Dysphagia Disorders Survey, Schedule for Oral Motor Assessment, Pre Speech Assessment Scale

Australian and New Zealand Standard Research Classifications (ANZSRC)

ANZSRC code: 110321, Rehabilitation and Therapy excl Physiotherapy, 60%
ANZSRC code: 111199, Nutrition and Dietetics not elsewhere classified, 20%
ANZSRC code: 111403, Paediatrics, 20%

Fields of Research (FoR) Classification

FoR code:1103, Clinical Sciences, 60%
FoR code: 1111, Nutrition and Dietetics, 20%
FoR code: 1114, Paediatrics and Reproductive Medicine, 20%

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List of Abbreviations used in the thesis

BAMF-OMD	Brief Assessment of Motor Function: Oral Motor Deglutition Scale
BMI	Body Mass Index
ca	Corrected age
CP	Cerebral Palsy
CFCS	Communication Function Classification System
CI	Confidence Interval
COSMIN	Consensus-based Standards for the Selection of Measurement Instruments
CPFQ	Cerebral Palsy Child Feeding Questionnaire
CRP	Centre for the Rehabilitation of the Paralysed
DDS	Dysphagia Disorders Survey
EAR	Estimated Average Requirement
EDACS	Eating and Drinking Ability Classification System
FBS	Feeding Behaviour Scale
FEES	Flexible endoscopic evaluation of swallowing
FFAm	Functional Feeding Assessment, modified
GMFCS	Gross Motor Function Classification System
GMFM	Gross Motor Function Measure
GNPA	Growth Nutrition and Physical Activity (study)
GVA	Gisel Video Assessment
ICF	International Classification of Functioning
kJ	Kilojoules
LRTI	Lower respiratory tract infection
MACS	Manual Ability Classification System
NHMRC	National Health and Medical Research Council (Australia)
OPD	Oropharyngeal dysphagia
OR	Odds Ratio
PEDI	Pediatric Evaluation of Disability Inventory
PSAS	Pre Speech Assessment Scale
SD	Standard Deviation
SOMA	Schedule for Oral Motor Assessment
TAGS	Tests in the Absence of a Gold Standard
TD	Typically development
VFSS	Videofluoroscopic Swallow Study

Chapter 1: Introduction, Thesis Outline and Aims

1.1. Introduction

Feeding and swallowing difficulties, or oropharyngeal dysphagia (OPD), are commonly occurring in children with cerebral palsy (CP)[1,2] and place them at risk of prolonged and stressful mealtimes, poor dietary intake and nutritional status, and compromised respiratory health.[3-5] Optimal nutritional status forms a critical foundation for general health and well-being across the lifespan; with compromised nutritional status influencing children's mood and irritability, muscle spasticity, wound healing, peripheral circulation, and immune response.[6] Both OPD and tube feeding have been demonstrated as independent risk factors for increased premature mortality in individuals with CP,[7-10] with respiratory related mortality the leading cause of premature mortality.[11,12] Cerebral palsy is the most common cause of physical disability in childhood, estimated at 2 per 1000 live born infants within Australia.[13] It is a group of disorders of movement, posture, or motor function, which are permanent but not unchanging, and due to a nonprogressive lesion to the immature brain.[13,14]

1.1.1 Cerebral Palsy

Cerebral palsy describes a clinical presentation rather than a specific aetiology or pathology.[15] As such, CP encompasses an heterogeneous group of individuals, varying by functional motor severity, motor type and distribution, and comorbidities (vision, hearing, speech, intellectual function, and epilepsy).[13,16] The direct cause of CP often remains unidentified, but of known events there are marked differences in the type and timing of the neurological lesion.[13] Based on the Australian CP Register Report, 94.4% of CP was acquired pre/ perinatally, with approximately 45% of cases born preterm,[13] which is reflective of the profile in most western developed countries.[17] In low-resource settings, however, it is proposed that this pattern is reversed, with most cases attributed to inadequate birthing practices and postnatal factors (kernicterus, meningitis, cerebral malaria).[18-20]

The type and timing of neurological lesion has been associated with the clinical picture of CP, with regards to motor type/ distribution and motor severity.[21] Depending on the location and extent of the neurological lesion, children's motor type may be described as spasticity, dyskinesia (athetosis and dystonia), ataxia, or hypotonia (Table 1).[22-24]

Furthermore, the number of limbs involved may also differ between individuals depending on the location of the lesion (unilateral vs bilateral, 1-4 limbs).[24] In the systematic review by Krägeloh-Mann & Horber, periventricular white matter lesions were the most common brain lesion in CP, and almost always associated with preterm births, and commonly mild bilateral spasticity of the lower limbs.[21] Grey matter lesions were most frequently identified in term born children, and associated with severe bilateral spasticity and dyskinetic CP. Brain maldevelopments, while infrequent, tended to be related to term births and severe forms of CP.[21]

Table 1. Classification of Cerebral Palsy Subtypes

Classification of CP subtypes		**All CP subtypes have in common an abnormal pattern of movement and posture. Additional features by subtype:**
Spastic CP	Bilateral spastic Unilateral spastic	Increased tone. Pathological reflexes: - Increased reflexes eg, hyperreflexia. - Pyramidal signs eg, Babinski response. Resulting in abnormal pattern of movement and posture.
Dyskinetic CP	Dystonic Choreo-athetotic	Involuntary, uncontrolled, recurring, occasionally stereotyped movements, primitive reflex patterns predominate, muscle tone is varying.
Ataxic CP		Loss of orderly muscular coordination, so that movements are performed with abnormal force.
Hypotonic		Decreased resistance to passive movement.

Reproduced from *Surveillence of Cerebral Palsy in Europe,* http://www.scpenetwork.eu/[24] and Sanger[22] for hypotonia definition.

The functional limitations associated with a diagnosis of CP also vary, ranging from mild impairments, resulting in minimal limitations to daily activities; to significant restrictions to the individual's daily activities and participation. To facilitate the standardised description of the gross motor function of an individual with CP, the Gross Motor Function Classification System (GMFCS) is increasingly being used (Table 2).[25] Children are classified to 1 of 5 levels (I being the greatest level of function, V being the poorest function) based on self-initiated movements in sitting, standing and walking. Owing to its excellent psychometrics and prognostic ability, the GMFCS has become a universally used language between clinicians and researchers working in the field of CP.

Table 2. Gross Motor Function Classification System

GMFCS I. Children walk indoors and outdoors, and climb stairs without limitation. Children perform gross motor skills including running and jumping but speed, balance and coordination are impaired.
GMFCS II. Children walk indoors and outdoors, and climb stairs holding onto a railing but experience limitations walking on uneven surfaces and inclines and walking in crowds or confined spaces. Children have at best only minimal ability to perform gross motor skills such as running and jumping.
GMFCS III. Children walk indoors or outdoors on a level surface with an assistive mobility device. Children may climb stairs holding onto a railing. Children may propel a wheelchair manually or are transported when travelling for long distances or outdoors on uneven terrain.
GMFCS IV. Children may continue to walk for short distances on a walker or rely more on wheeled mobility at home, school and in the community. Children may achieve self-mobility using a power wheelchair.
GMFCS V. Physical impairments restrict voluntary control of movement and the ability to maintain antigravity head and trunk postures. All areas of motor function are limited. Children have no means of independent mobility and are transported.

Reproduced from Palisano et al (1997)[25]

1.1.2 Oropharyngeal Dysphagia in Cerebral Palsy

In addition to influencing skeletal muscles, the neurological lesion associated with CP may impact on muscle function of the jaw, cheeks, lips, tongue, palate and pharynx.[26] This may manifest functionally as difficulties with controlling saliva, eating, drinking, swallowing and speaking. The neural control of eating and drinking has been well defined, as a complex process involving the brainstem in addition to widely distributed, bilateral and multifocal cortical and subcortical central structures.[27] The process of eating and drinking is typically described in 3-4 distinct but overlapping phases, including the oral phase (sometimes further divided into the oral-preparatory and oral propulsive phases), the pharyngeal, and oesophageal phases of the swallow, as represented visually in Figure 1.[28] While there is generally agreement in the literature with regards to the afore-mentioned phases of the swallow, there is little consensus regarding the terms used to describe feeding impairments or mealtime activity limitations, or the construct parameters. The terms OPD, *feeding/ deglutition disorder* and *oral motor dysfunction* have been used frequently, but have variably included delayed and dysfunctional feeding; motor, sensory and/ or behavioural feeding difficulties; oropharyngeal impairments and those to self-feeding. This variability in the domains encompassed by these terms has in part lead to variability in results of studies of OPD. This thesis focuses on OPD in preschool children

with CP, defined as impairment to one or more phases associated with eating, drinking, or controlling saliva.

1.1.2.1 Oral-preparatory Phase

The oral-preparatory phase is initiated when food or fluid is taken into the mouth, and involves tasks necessary in bolus formation: including sucking, biting, munching and chewing. Food and fluid are contained in the oral cavity surrounded by the upper dental arch and closure of the lips. Posterior leakage of the fluid bolus is prevented by contact between the soft palate and tongue, however this contact is not maintained during the processing of the solid bolus. The oral (propulsive) phase involves the backward propulsion of the bolus, by the tongue gradually expanding its contact with the hard palate posteriorly, to initiate the pharyngeal swallow.[28,29] The duration and movements necessary for the oral phases differ depending on the child's age, textures ingested, and the utensils.[29,30] When defining the swallow stages for solid foods, Matsuo and Palmer advocate the use of the *Process Model of Feeding*, because of the overlap between the phases described in the *Four Stage Model* for fluids.[28] The *Process Model* further divides the oral-preparatory phase into *Stage I Transport*, and *Food Processing*. During *Stage I Transport*, the food is first ingested and moved onto the lateral occlusal surfaces of the teeth. *Food Processing* involves the mastication of solids to an optimal consistency for swallowing. *Stage II Transport* is equivalent to the oral propulsive phase of the four stage model for fluids (backward propulsion of the bolus).

1.1.2.2 Pharyngeal Phase

The pharyngeal phase of the swallow describes the passage of food or fluid boluses through the pharynx.[28] On initiation of the pharyngeal phase, the soft palate elevates to seal the nasopharynx to prevent nasal regurgitation. The tongue base retracts, propelling the bolus posteriorly against the pharyngeal walls followed by the pharyngeal constrictor muscles contracting to squeeze the bolus downward. To ensure airway safety during bolus passage, respiration ceases momentarily (deglutition apnoea), the vocal folds close, the arytenoids tilt forward to contact the base of the epiglottis, the larynx elevates under the base of the tongue, and the epiglottis inverts to seal the laryngeal vestibule. The opening of the upper oesophageal sphincter is facilitated through the relaxation of the cricopharyngeous muscle, contraction of the suprahyoid and thyrohyoid muscles, and the pressure of the descending bolus.[28] The oesophageal phase is the final phase of the

swallow, which begins as the bolus moves through the upper oesophageal sphincter, to be transported via automatic peristaltic waves to the stomach.[29]

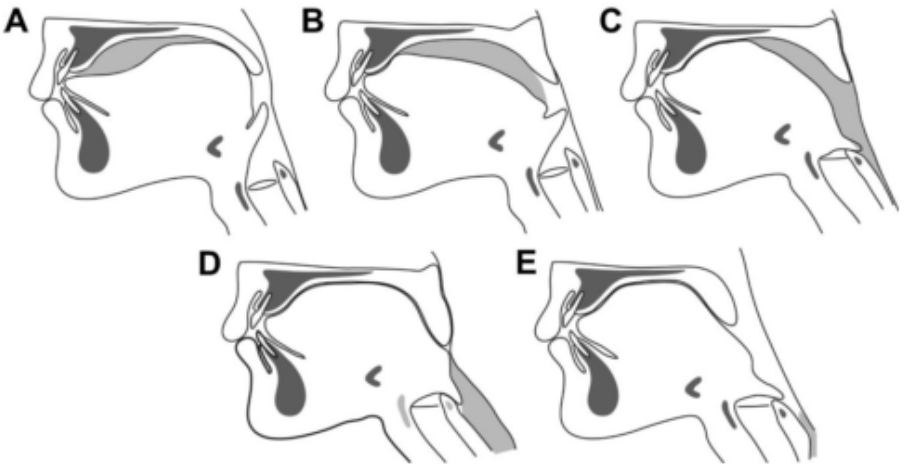

Figure 1. Diagram of the Swallowing of a Bolus

Figure reprinted from Physical Medicine and Rehabilitation Clinics of North America, Vol 19, Matsuo K & Palmer JB, Anatomy and Physiology of Feeding and Swallowing – Normal and Abnormal, Pages No. 691-707, Copyright 2008, with permission from Elsevier. Drawings based on a videofluorographic recording, depicting the normal swallow of a fluid bolus. Note, that in swallowing a solid bolus, the oral propulsive and pharyngeal phases (C-D) are more discrete.

1.1.2.3 Oropharyngeal Dysphagia in Cerebral Palsy

Oropharyngeal dysphagia in children with CP tends to be characterised by motor impairments of the oral and pharyngeal phases of the swallow, although children may also have co-occurring limitations to sensory, cognitive, behavioural (including motivational, such as appetite), and structural domains. Little has been written regarding the specific neurological basis of OPD in children with CP. A recent review, however, hypothesised that the hypoxic-ischemic pathology associated with CP may correspond to predictable patterns of OPD. Children with cortical lesions are more likely to experience difficulty with the volitional aspects associated with the oral phase of the swallow, whereas those with subcortical and basal ganglia lesions having frequent pharyngeal impairments in addition to those of the oral phase. Children with brainstem lesions may have difficulty coordinating the swallow-respiratory cycles, which may increase their risk of aspiration.[27] The range of impairments associated with a diagnosis of CP include altered tone, weak or reduced movement of the oropharyngeal structures (lips, cheeks, tongue, jaw and pharynx), discoordination of movement (including that needed to coordinate the swallow with breathing), and an increase or persistence of oral reflexes (such as tongue thrust, gag, or

bite reflex).[5] These impairments may impact on 1 or more ingestion functions of the oral phase, including visual attention to the bolus, receiving and maintaining the bolus in the oral cavity, moving the bolus through the mouth to process it for swallowing, and propulsion of the bolus posteriorly. They may also influence the safety of bolus passage through the pharynx, including premature spillage of the bolus, delayed initiation of the swallow, inadequate closure of the laryngeal vestibule, or inadequate bolus clearance/ residue after the swallow. These factors place children at risk of oropharyngeal aspiration, or food or fluid entering the trachea below the vocal folds,[31] which is a commonly cited risk factor for recurrent pneumonia.[32] The ingestion functions of eating or drinking, and potentially associated difficulties for a child with CP, are summarised in Table 3.

1.1.2.4 Limitations to Food and Fluid Textures Associated with Oropharyngeal Dysphagia

Oropharyngeal sensorimotor impairments may influence the range of food and fluid textures children with CP can safely and efficiently manage. Food and fluid textures are classified using standardised terminology, according to similarities in their physical properties, such as firmness and flow rate.[33] Modifications to food/ fluid textures may be recommended to address mealtime safety or efficiency, or to encourage development of oral sensorimotor skills.[34] There is generally consensus that risk of aspiration is increased when taking thin fluids.[35,36] Chewable foods may present a choking hazard if poorly masticated, and they are less efficiently consumed compared to purees in children with CP and those with typical development.[37] Foods typically introduced to children at a younger age, such as purees, are processed on the midline with a suckle feeding pattern, while more complex textures, such as chewable foods, require tongue lateralisation and separation of movement (of lip, tongue and jaw function).[38]

1.1.2.5 Developmental Changes to Feeding Skills

Children's feeding skills undergo a series of important changes through infancy and the preschool years as part of typical development, from suckle feeding in infancy, to the rapid oropharyngeal skill changes and encephalization during transitional feeding (4 to 36 months),[39] and finally a period of skill consolidation (3 to 6 years).[39,40] The range of food textures children can safely and efficiently ingest are gradually expanded, and by 18 to 24 months, children can typically chew and swallow firm chewable foods, and those with dual textures (eg, juicy fruits).[41] The complexity of chewable foods children can safely manage corresponds to their pattern of chewing; which moves from a rhythmic phasic bite reflex in

infancy, graduating into more integrated chewing patterns with vertical single-plane movements, and finally multi-plane rotary chewing by around 24 to 36 months.[39,42] The fluid utensils children use also progress through these early years, and influence how independently, efficiently and safely children manage drinks. Many children may continue to use cups with lids until 20 months, before regularly drinking from an open cup between 24 to 36 months.[41] Each of these periods of feeding development may present varied challenges for a child with CP, as more complex textures, greater volumes of intake, utensils and mealtime independence/ routines place additional requirements on the child's oral sensorimotor, swallow-respiratory and cognitive systems.

1.1.2 Assessment of Oropharyngeal Dysphagia

It is necessary to comprehensively assess children's OPD in order to: (1) achieve safe mealtimes, including eliminating or minimising risk of aspiration, (2) ensure adequate and efficient dietary intake (including energy, micronutrients and hydration), (3) promote age appropriate or optimal oropharyngeal sensorimotor function. This may occur through a combination of feeding recommendations and medical management. Medical management may include decision making surrounding health consequences associated with OPD, such as introduction of supplementary nutrition (including the insertion of a gastrostomy feeding tube), cessation or modification of oral feeding in instances of aspiration or recurrent pneumonia, or prescription of medication/ fundoplication for gastro-oesophageal reflux. Clinicians working in the field of dysphagia typically adopt a number of treatment strategies: including (1) direct: oropharyngeal sensorimotor treatments to target the specific physiological limitations or impairments (techniques targeting oral-motor and swallow function, as well as non-nutritive stimulation); and (2) indirect: education for the primary caregiver regarding the feeding environment or mealtime routines; or adjustment to the child's food/ fluid textures, utensils, feeding patterns, or positioning.[43-47] While this thesis aims to provide information to assist in planning of OPD interventions, a detailed exploration of intervention was not in the scope of this thesis.

Oropharyngeal dysphagia can be assessed using parent questionnaire, clinical assessment (standardised or informal), or instrumental assessments (most commonly a videofluoroscopic swallow study [VFSS] or flexible endoscopic evaluation of swallowing [FEES]).[5] Initially OPD is evaluated during a clinical feeding evaluation which would include review of medical and feeding history, the child's arousal level, oral sensorimotor evaluation, and mealtime assessment using a range of food/ fluid textures and utensils.

This initial clinical evaluation may be conducted using direct assessment by a speech pathologist or feeding clinician, as well as gathering information about the child's typical mealtime performance from parent questionnaire. Results of the clinical feeding evaluation are used to plan management goals as well as indicating the need for instrumental assessment.[48] Instrumental assessments usually focus on specific aspects of swallowing, such as identification of oropharyngeal aspiration,[48-50] that can only be inferred from a clinical feeding assessment.[5,51] In isolation, however, instrumental assessments do not provide a holistic view of the child's OPD. Clinical assessments can be more subjective, owing to their reliance on the knowledge and experience of the clinician conducting the assessment, however they provide valuable information regarding a child's oral sensorimotor and swallowing skills, behaviour and interaction during feeding in a mealtime context. The use of formal measures with strong psychometric properties to assess OPD can improve the objectivity of clinical feeding evaluations. There is no single measure considered the gold standard to evaluate OPD in children with CP.[52]

1.1.3 Prevalence of Oropharyngeal Dysphagia and Risk Factors

OPD prevalence estimates in the literature have varied considerably,[1,2,10,26,53-62] with estimates ranging from as low as 19% in a large register sample,[10] up to 99% in a sample of children with moderate to severe CP.[1] Generalisability of the prevalence and patterns of OPD is limited by variability in study methodology, sampling bias, and inconsistency in the case definition of OPD. Many studies have based the prevalence of OPD on parent report or informal methods, and samples have generally been limited to children with more severe gross motor impairments[1,53,55,57] and across a broad age range.[1,26,53,55-61] The use of opportunistic sampling methods, such as recruitment through hospitals, rehabilitation centres or special schools are a significant factor influencing the skewed sample characteristics. The literature is also limited to exploration of OPD in children from primarily Caucasian ethnicity and within a high-resource context. With 80% of the global burden of CP estimated to be in low-resource countries (non-Caucasian),[18] an understanding of OPD in this context is critical in understanding the broader prevalence and patterns of OPD.

A number of intrinsic and extrinsic factors have been associated with the presence of OPD, many of which are interrelated. In a cross-sectional study of children with childhood impairments (n=343, of which 96% had CP), children with OPD were significantly more likely to attend special schools, and more likely to have comorbidities such as epilepsy, intellectual impairment, visual impairment, and difficulty with speech.[1,56]

A number of studies have also supported the positive association between poorer gross motor function and presence of OPD.[1,2,10,26,55,56] Consistent with this association between poorer gross motor function and OPD (with a strong association between GMFCS and motor type),[63] children with unilateral spasticity were also less likely to to have OPD than other motor types.[56,62] In addition to the relationship between motor type and GMFCS,[63] the relationship between gross motor functional severity and OPD may reflect the importance of trunk and head stability and alignment for feeding,[55,64,65] but may also be a marker of the severity of the neurological lesion.

Table 3. Oropharyngeal Dysphagia in Children with Cerebral Palsy

Ingestion function	Component tasks	Potential difficulties in CP
Orienting to the bolus	Visual and cognitive attention Head movement towards oncoming bolus	Comorbidities (visual, hearing, cognition) Head and trunk instability Interest/ appetite/ pain
Receiving the bolus (spoon)	Lip clearing/ downward movement Jaw stability	Inability to clear spoon with lips Lack of graded jaw opening Inability to maintain stable jaw Tonic bite reflex
Receiving the bolus (bite)	Jaw strength and stability	Inadequate strength to break through food (resulting in sucking/ tearing) Phasic/ tonic bite reflex on food Overflow movement in body Inappropriate bite size (too small/ large (mouth stuffing))
Receiving the bolus (bottle, cup or straw)	Lip movement to strip teat/ suck Jaw stability Tongue action to strip or suck	Fluid loss during receiving the bolus Inefficient/ slow sucking Inappropriate bolus size (small single sips only or inability to control speed of flow)
Maintaining the bolus in the oral cavity	Lip closure Velopharyngeal closure Tongue position to prevent posterior spillage Maintaining bolus cohesion	Food/ fluid loss through lips Nasopharyngeal backflow/ regurgitation Tongue thrusting/ exaggerated tongue protraction
Oral transport	Tongue function Cheek strength	Oral residue post swallow Food in anterior/ lateral sulci Increased oral transit time Head extension to use gravity for bolus trasport

Chewing	Tongue lateralisation Cheek strength Jaw strength and control	Inability to lateralise food to teeth Mashing foods on roof of mouth Inadequately chewed food (strength or sensory input)
Swallow	Strong propulsion by tongue base Swallow-breath synchrony Velopharyngeal closure Closure of airway Contraction of pharynx Coordinating consecutive swallows	Premature spillage Delayed initiation of swallow Nasopharyngeal backflow/ regurgitation Inadequate closure of larynx Penetration or aspiration Pharyngeal residue post swallow (eg, in the valleculae, pyriform sinuses, posterior pharyngeal wall) Difficulty with consecutive swallows
Gastro-eosophageal	Opening of upper oesophageal sphincter Bolus passage to stomach through peristalsis	Incomplete pharyngeal bolus clearance with pyriform sinus residue Gastro-oesophageal reflux

Adapted from Arvedson 2013[5], Sheppard 2003[66], Morris 2000[67]

1.2 Frameworks and Definitions for Oropharyngeal Dysphagia

1.2.1 Theoretical Framework and Models

Oropharyngeal dysphagia can be considered holistically using the body structure and function, activity and participation model of the *International Classification of Functioning, Disability and Health* (Figure 2).[68] This theoretical framework of disability and functioning allows a broader understanding of OPD, in the context of an individual's functioning in society, and considering personal and environmental factors, shifting the focus from cause to impact.[68] Diagnosis alone does not predict service needs or functional outcomes of an individual, therefore this study considers the individual oral structures (eg, tongue, lips, jaw) during ingestion functions (eg, biting, sucking, chewing), within the activities of eating, drinking and controlling saliva. The broader aspects of the activities of eating and drinking are outside the scope of the thesis.

Mealtime assessments conducted as part of this doctoral research were in a clinical setting following a standardised protocol (Appendix 20). The naturalistic components of the mealtime, such as the child's typical utensils, preferred foods, and feeding position were retained as far as possible. This recognises the critical role of environmental and personal

factors on body functions and the performance of activities. Use of parent questionnaires also allowed the collection of information specific to mealtimes in the home environment.

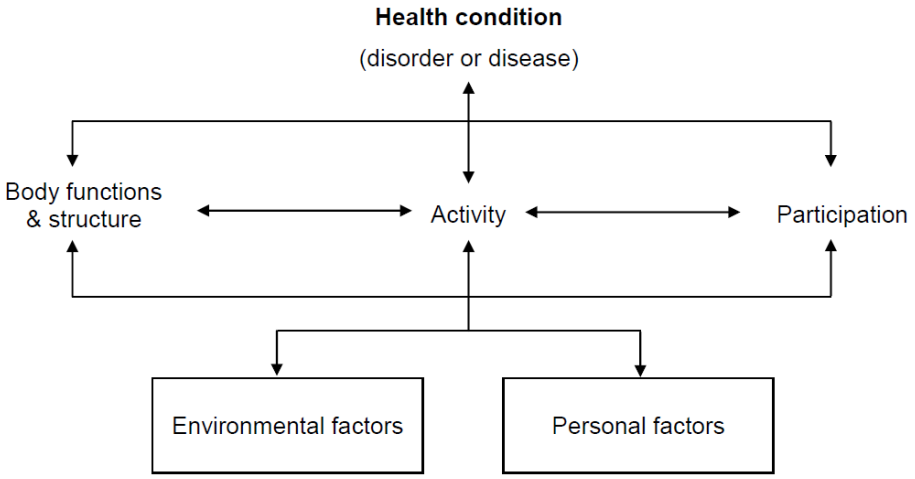

Figure 2. International Classification of Functioning, Disability and Health (ICF) Model
Reproduced from the World Health Organization http://www.who.int/classifications/icf/en/[68]

1.2.2 Operational Definitions of Study Variables

This doctoral research explores the prevalence and patterns of OPD and its relationship to dietary intake, nutritional status and gross motor function. These variables are shown conceptually in Figure 3, and defined as follows:

Oropharyngeal Dysphagia: impairment to any component of the oral and/ or pharyngeal phases associated with eating, drinking or controlling saliva.
Feeding: the activities of eating and drinking.
Eating: Carrying out the coordinated tasks and actions of eating food that has been served, bringing it to the mouth and consuming it in culturally acceptable ways, cutting or breaking food into pieces, opening bottles and cans, using eating implements, having meals, feasting or dining.[68]
Drinking: Taking hold of a drink, bringing it to the mouth and consuming the drink in culturally acceptable ways, mixing, stirring and pouring liquids for drinking, opening bottles and cans, drinking through a straw or drinking running water such as from a tap or a spring, feeding from the breast.[68]
Ingestion functions: Functions related to taking in and manipulating solids or liquids through the mouth into the body.[68]

Gross motor function: Abilities and limitations to tasks such as sitting, standing, walking and climbing stairs, in the contexts of home, school and community.[69]

Dietary intake: Amounts and types of food and drink consumed, including dietary supplements. Provides an indication of whether nutrient requirements are being met.[70]

Nutritional status: Indication of the adequacy of an individual's dietary intake to maintain healthy body size and function with reference to a population, based on the individual's age and gender. Nutritional status is frequently measured using anthropometric measurements, biochemical tests, clinical indicators and dietary assessments.[70]

Growth: Indicator of nutritional status in children, most commonly measured by height for age, weight for age and weight for height.[70]

Respiratory health: Includes pneumonitis from aspiration of food and vomit (J69.0).[71]

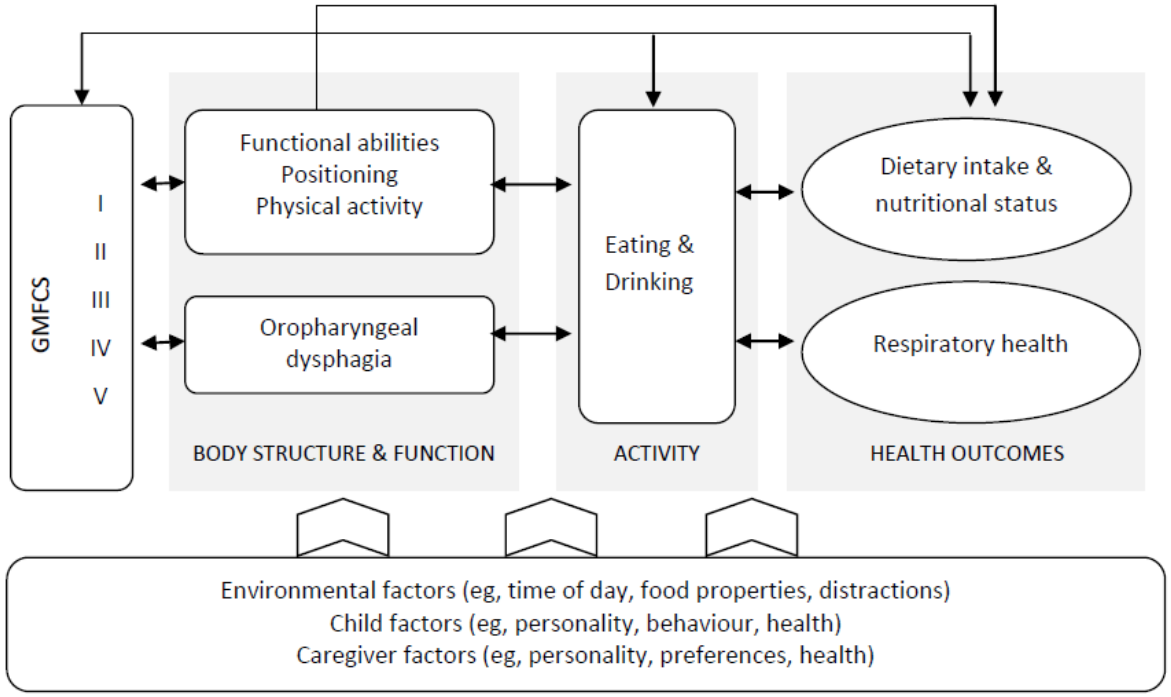

Figure 3. Theoretical Relationship Between Doctoral Research Study Variables

1.3 Thesis Outline

To date there has been limited analysis of the prevalence and patterns of OPD in preschool children with CP, particularly in a representative sample and evaluated using valid and reliable measures. This data would form a necessary foundation for research in the field, and allow the interpretation and generalisation of the findings to other samples. The prevalence and patterns of OPD in young children with CP are critical to understand in order to plan and provide earlier detection and interventions, thus minimising the potential

health impacts of OPD, including poor nutrition and compromised chest status. As such, this doctoral research program investigated the relationship between OPD, dietary intake, nutritional status and gross motor function in preschool children with CP across 2 age points, 18 to 24 months and 36 months (corrected age, ca). It explored these relationships in a population-based sample across the full spectrum of motor severity (GMFCS I-V) in 4 substudies:

1. Systematic review of OPD measures, and testing of validity and reproducibility:
    a. Clinimetric review of measure psychometrics and clinical utility.
    b. Discriminative validity with typically developing (TD) reference sample.
    c. Convergent validity between 3 direct OPD measures.
    d. Reproducibility (test-retest, intrarater, interrater reliability and agreement).
    e. Agreement between direct OPD assessment and parent report.
2. Cross-sectional study of children aged 18 to 36 months:
    a. Overall prevalence of OPD, subtypes and association with gross motor function.
    b. Functional feeding impairments on food and fluid textures.
    c. Oral phase impairments.
    d. Pharyngeal phase impairments.
3. Longitudinal study of children with CP between 18 to 24 months and 36 months ca.
4. Oropharyngeal dysphagia and its relationship to GMFCS in a high-resource and low-resource country.

1.4 Aims and Hypotheses

The broad aim of this doctoral research was to determine the prevalence and patterns of OPD in preschool children with CP aged 18 to 36 months ca; and its relationship to dietary intake, nutritional status and gross motor function. The specific aims of the study are described according to each substudy.

1.4.1 Substudy 1: Validity and Reproducibility of Oropharyngeal Dysphagia Measures

1^{A} Systematically review the literature determining the clinimetric properties of measures of OPD in preschool children with CP.

1^{B} Test the convergent validity of the *Schedule for Oral Motor Assessment* (SOMA), *Dysphagia Disorders Survey* (DDS) and *Pre Speech Assessment Scale* (PSAS) by triangulating the 3 measures.

1^{C} Determine the discriminative validity of the SOMA, DDS, PSAS and clinical signs suggestive of pharyngeal phase impairment by comparison to a TD reference sample.

1^{D} Determine the reproducibility (test-retest, intrarater and interrater reliability, and percentage agreement) of the SOMA, DDS, PSAS and clinical signs suggestive of pharyngeal phase impairment.

1^{E} Determine the agreement between parent-reported OPD prevalence and severity, and directly assessed OPD (for oral phase, pharyngeal phase and textures).

H^{1A} The SOMA and DDS will be the most valid and reproducible direct clinical measures of OPD in preschool children with CP. The PSAS will have the best clinical utility.

H^{1B} The prevalence of OPD detected on the PSAS will be equivalent to that on the DDS, but greater than that on the SOMA.

H^{1C} The specificity will be highest for the SOMA, but lower for the DDS and PSAS.

H^{1D} There will be excellent reproducibility (test-retest, intrarater, interrater) for all OPD measures (including overall, the SOMA, DDS, PSAS, clinical signs).

H^{1E} Parents will detect clinically significant/ overt OPD but will underdetect mild OPD.

Rationale: In order to improve both clinical screening for OPD and progress research in the field, we need to understand how the more frequently used measures perform with regards to their validity and reproducibility. In addition, understanding whether these measures are all exploring the same construct will assist in better measure selection. Parent report has been used extensively in research on OPD to date, although there is a limited understanding of the accuracy of this as a proxy for a direct objective assessment. If parent report is shown to be accurate, it may be a more economical and feasible method of clinical screening.

1.4.2 Substudy 2: Cross-Sectional Study of Oropharyngeal Dysphagia in Children Aged 18 to 36 Months

2^{A} Determine the prevalence of OPD and its subtypes (impaired saliva control, oral phase impairment, and pharyngeal phase impairment) in a population-based sample of children with CP at 18 to 36 months.

2^{B} Describe the nature of OPD subtypes (oral phase and pharyngeal phase) in children with CP at 18 to 36 months.

2^{C} Explore the nature of the relationship between OPD and gross motor function (according to GMFCS levels, motor type, and distribution).

H^{2A} The prevalence of feeding difficulties will be lower than that reported in the literature (based on a more representative sample of children with CP from all motor severities).

H^{2B} (a) Children with ambulatory CP (GMFCS I-II) will have only delayed feeding skills, whereas children with nonambulatory CP (GMFCS IV-V) will have delayed and disordered skills.

(b) Clinical signs suggestive of pharyngeal phase impairment will be present across GMFCS levels. Cough will be the most frequent sign in children from GMFCS I-II.

H^{2C} There will be a negative relationship between OPD prevalence and severity, and gross motor function in children with CP aged 18 to 36 months.

Rationale: Most of the literature to date has relied on parent-reported OPD, and explored prevalence and patterns in a sample of school-aged children and those with moderate to severe CP. There is a paucity of data regarding early feeding abilities in children with CP, or the skills of children with ambulatory CP (ie, GMFCS I-II). Early intervention for children with CP is critical, both from a motor learning perspective, as well as to prevent secondary consequences such as poor growth and compromised respiratory status. Children aged 18 to 36 months are typically eating a full range of table foods, thus for a child with CP this may be an age when progress begins to lag behind their peers. It is not known if young children with ambulatory CP also have OPD, and if so, whether the patterns differ from that of children with nonambulatory CP. There have only been 3 studies, to our knowledge, of OPD in ambulatory children with CP, and these have been conducted in school aged samples.[38,56,72] Understanding these patterns will help with improved screening and management planning, but will also assist in designing studies to evaluate the response of specific impairments to oral sensorimotor treatments.

1.4.3 Substudy 3: Longitudinal Study of Oropharyngeal Dysphagia in Children with Cerebral Palsy Between 18 to 24 Months and at 36 Months

3^{A} Determine the change in OPD prevalence and severity between 2 critical time points, 18 to 24 months and at 36 months ca.

3^{B} Determine the relationship between change in OPD prevalence and severity and change in GMFCS level; and examine other potential risk factors for OPD (motor type and distribution, preterm status, epilepsy, gender, age, socio-economic status).

3^{C} Longitudinally examine the association between OPD at 18 to 24 months and health outcomes (nutritional status, nutritional interventions, respiratory health and parent stress) at 36 months.

H^{3A} OPD prevalence will be lower at 36 months compared to 18 to 24 months, owing to a later maturation of oropharyngeal feeding skills in children with CP.

H^{3B} GMFCS will be more strongly associated with OPD prevalence at assessment 2 (36 months) compared to assessment 1 (18 to 24 months).

H^{3C} OPD severity at 18 to 24 months will be strongly associated with poor growth, poor nutritional status, and introduction of nutritional interventions at 36 months.

Rationale: There have been no studies to our knowledge which have explored the changes to OPD prevalence or severity in preschool children aged 18 to 36 months with CP using longitudinal methods. These age bands (18 to 24 and 36 months) represent an important period of skill development, with 18 months representing an age when children can typically manage the full range of food textures, and 36 months the beginning of a period of skill consolidation. Thus this period may present additional challenges for a child with CP, representing an important stage for early detection and intervention of OPD for children at risk of nutritional compromise. Knowledge of risk factors associated with OPD will provide foundational data to fill a significant knowledge gap. It will also contribute pilot data to begin to predict the patterns of OPD which are significant with regards to health outcomes, such as growth and body composition, respiratory health, and parent stress in mealtimes.

1.4.4 Substudy 4: Oropharyngeal Dysphagia in Children with Cerebral Palsy Aged 18 to 36 months Residing in a Low-Resource Country

4^{A} Determine the prevalence of OPD and its subtypes (impaired saliva control, oral phase impairment, and pharyngeal phase impairment) in children with CP residing in a low-resource country, at 18 to 36 months.

4^{B} Describe the risk factors and nature of OPD in children born in Bangladesh with CP aged 18 to 36 months.

4^{C} Compare the prevalence, severity and risk factors for OPD in children born in Bangladesh to that in children born in Australia with CP.

H^{4A} The prevalence and severity of OPD in Bangladesh will be greater than that reported in the literature.

H^{4B} The prevalence and severity of OPD and its subtypes will have a strong positive relationship with gross motor function in both countries.

H^{4C} The prevalence and severity of OPD in children with CP from Bangladesh, when stratified for GMFCS level, will be greater than that in the Australian sample.

Rationale: The rationale for the inclusion of this forth substudy is 3-fold. (1) There is a paucity of research on OPD in CP, but much of the research that exists is targeted at describing OPD in the context of Western high-resource countries. It has been estimated that 80% of the global prevalence of CP may be in low-resource countries alone.[18] In low-resource settings where health resources are already scarce, the need for information to assist in service planning and prioritisation is even more critical. (2) The inclusion of this sample allows the testing of the hypothesis, that there is a strong relationship between OPD and gross motor function. This strengthens the conclusions drawn from this thesis, and allows generalisability of the findings to different populations. (3) There was an opportunistic aspect to this substudy, as the candidate has had significant previous work experience with cerebral palsy in Bangladesh.

1.5 Format of Thesis

This thesis is a compilation of 10 papers (published or submitted to peer-reviewed journals). Chapter 2 presents the published clinimetric review of the measures of OPD, proposing those with the best psychometric properties and clinical utility for use in the doctoral research. Chapter 3 contains a description of the methods, by way of inclusion of the published protocol paper. It also discusses the literature relating to prevalence of OPD, and gaps and limitations in the current research. New literature arising since the publication of this protocol will be presented in this chapter.

The results of the cross-sectional research related to the Queensland cohort are presented in chapters 4 to 8, with each chapter including a brief introduction, a peer-reviewed article or manuscript, and chapter summary. Chapter 4 presents the overall prevalence data for OPD and its subtypes (oral phase, pharyngeal phase, saliva control),

and its relationship with gross motor function, motor type, and distribution. Chapter 5 discusses the findings of the study which tested the psychometric properties of the SOMA, DDS, and PSAS. In this chapter, data from the children with TD are analysed with reference to children with CP. Chapter 6 investigates the functional feeding skills of children, by analysing the range of food and fluid textures included in children's diets by their parents, the proportion of textures habitually consumed, and relationship to dietary intake. Chapter 7 explores the specific oral phase impairments, and chapter 8 the clinical signs associated with pharyngeal phase impairments, both presented according to children's gross motor function.

Chapter 9 presents the findings from the longitudinal study, exploring changes in OPD between 2 critical age points, 18 to 24 months and 36 months ca. This chapter discusses change in prevalence between assessments, OPD risk factors, and association with health outcomes at 36 months. The last 2 results papers, in Chapter 10, provide a summary of the cross-sectional study in Bangladesh, which sought to explore differences in the motor patterns and OPD in an economically and geographically contrasting context. This chapter presents a more detailed introduction to this substudy, the results of the substudy as a peer-reviewed article and manuscript (under review), and a chapter summary. Finally, chapter 11 provides a general discussion, linking the various results papers related to the doctoral research. It will conclude by discussing the research and clinical implications of the doctoral research, followed by limitations of the overall study, and future research directions.

Chapter 2: Systematic Review of Measures of Oropharyngeal Dysphagia

Introduction to Chapter 2

This chapter includes the published article “Clinimetrics of Measures of Oropharyngeal Dysphagia for Preschool Children with Cerebral Palsy and Neurodevelopmental Disabilities: a Systematic Review”. The review was conducted to allow the selection of the most appropriate measures of OPD for preschool children with CP, the measurement of which is central to the doctoral thesis. As the field of feeding is relatively small, particularly as it relates to children with CP, the review was broadened to include measures appropriate for children with CP as well as neurodevelopmental disabilities.

Paper 1: Clinimetrics of Measures of Oropharyngeal Dysphagia for Preschool Children with Cerebral Palsy and Neurodevelopmental Disabilities: A Systematic Review

This paper was published in Developmental Medicine and Child Neurology (journal impact factor 3.292) and has been cited 16 times. It has been reproduced from Developmental Medicine and Child Neurology, Vol 54, Benfer KA, Weir KA, Bell KL, Ware RS, Davies P SW, Boyd RN, Clinimetrics of Measures of Oropharyngeal Dysphagia for Preschool Children with Cerebral Palsy, pages no. 784-795, Copyright 2014, © The Authors. Developmental Medicine & Child Neurology © 2012 Mac Keith Press, with permission from John Wiley and Sons.

Benfer KA, Weir KA, Boyd RN. Clinimetrics of measures of oropharyngeal dysphagia for preschool children with cerebral palsy and neurodevelopmental disabilities: a systematic review. *Dev Med Child Neurol*. 2012;54(9):784-95.

This paper was also presented as a free paper at the 6th Biennial Conference of the Australasian Academy of Cerebral Palsy and Developmental Medicine, May-June, Brisbane, Australia.

Benfer K.A., Weir K.A., Bell, K.L., Davies, P.S.W., Ware, R.S., Boyd R.N. A systematic review of the clinimetric properties of oral motor dysfunction measures for preschool children with cerebral palsy. *Dev Med Child Neurol*. 2012;54(Supp 5):20.

Clinimetrics of measures of oropharyngeal dysphagia for preschool children with cerebral palsy and neurodevelopmental disabilities: a systematic review

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PUBLICATION DATA
Accepted for publication 22nd February 2012.
Published online

ABBREVIATIONS

BAMF-OMD	Brief Assessment of Motor Function – Oral Motor Deglutition Scale
BASOFF	Behavioral Assessment Scale of Oral Functions in Feeding
DDS	Dysphagia Disorders Survey
FBS	Feeding Behaviour Scale
FFAm	Functional Feeding Assessment, modified
GVA	Gisel Video Assessment
OMAS	Oral Motor Assessment Scale
OPD	Oropharyngeal dysphagia
PSAS	Pre-Speech Assessment Scale
SOMA	Schedule for Oral Motor Assessment

AIM The aim of this study was to determine the psychometric properties and clinical utility of objective measures of oropharyngeal dysphagia (OPD) in children with cerebral palsy or neurodevelopmental disabilities aged 12 months to 5 years.
METHOD Five electronic databases were searched to identify measures of OPD. The Consensus-based Standards for the Selection of Measurement Instruments (COSMIN) Checklist was used to assess psychometric properties and a Modified *CanChild* Outcome Rating Form was used for clinical utility.
RESULTS Nine measures of OPD from 27 papers were assessed: the Brief Assessment of Motor Function – Oral Motor Deglutition Scale; the Behavioral Assessment Scale of Oral Functions in Feeding; the Dysphagia Disorders Survey; the Feeding Behaviour Scale; the Functional Feeding Assessment, modified; the Gisel Video Assessment; the Oral Motor Assessment Scale; the Pre-Speech Assessment Scale; and the Schedule for Oral Motor Assessment.
INTERPRETATION The Schedule for Oral Motor Assessment and the Functional Feeding Assessment, modified, proved to be the strongest measures based on published psychometric properties of validity and reliability. The Schedule for Oral Motor Assessment and the Dysphagia Disorders Survey were found to have the strongest clinical utility. Further studies to test the psychometric properties of existing measures, in particular predictive validity, responsiveness, and test–retest reliability, would be beneficial for selecting an appropriate measure for both clinical and research contexts.

Oropharyngeal dysphagia (OPD) is present in 90% of children with cerebral palsy (CP)[1] and is a major risk factor for morbidity and mortality in this population.[2] OPD leads to the inability to consume sufficient food and fluids safely and is associated with prolonged mealtimes, poor growth and nutrition, and respiratory consequences from oropharyngeal aspiration, all of which contribute to lowered health outcomes.[2,3] OPD encompasses impairment to any component of the oral-preparatory, oral (propulsive), and/or pharyngeal phases of swallowing that are associated with eating, drinking, or controlling saliva. The oral-preparatory phase is initiated when food is taken into the mouth, and involves tasks necessary in bolus formation, including sucking, munching, and chewing. The oral (propulsive) phase describes the posterior propulsion of the food bolus through the oral cavity. The pharyngeal phase is characterized by the passage of food or fluid through the pharynx and upper oesophageal sphincter to the oesophagus and the prevention of food entry into the airway.[4] The operational definition of OPD describes impairment to the ingestion functions (b510), defined in the International Classification of Functioning, Disability and Health (ICF) as 'functions related to taking in and manipulating solids or liquids through the mouth into the body'.[5]

According to the ICF, the activities of eating and drinking involve many aspects of upper limb function, social and cultural components, as well as ingestion functions.[5] These broader aspects of mealtime management contribute to a comprehensive mealtime assessment;[3,6,7] however, they are beyond the scope of this review. Children's oral sensorimotor skills develop rapidly through the first year of life, from a suckle pattern at birth during breast/bottle feeding through to manipulating more complex textures. Twelve months represents the age at which children can typically eat a range of food textures and drink from a cup or straw, and by 36 months they are expected to manage adult table foods.[8] Growth in the early years is critical, owing to increased energy needs, so the evaluation of oral sensorimotor and swallowing skills and careful nutritional monitoring are indicated in at-risk populations such as those with CP.

Initially, OPD is evaluated with clinical measures, and the results are used to plan management goals and indi-

DOI: 10.1111/j.1469-8749.2012.04302.x

cate the need for instrumental assessment.[9] Instrumental assessments usually focus on specific aspects of swallowing, such as identification of oropharyngeal aspiration,[6,9,10] that are poorly detected in a clinical feeding assessment,[11] but do not provide a holistic view of the child's feeding. Clinical evaluations can be more subjective, because of their reliance on the knowledge and experience of the professional conducting the assessment; however, they provide valuable information regarding a child's oral sensorimotor and swallowing skills, behaviour, and interaction during feeding in a mealtime context.

The use of formal clinical measures with strong psychometric properties to assess OPD can improve the objectivity of clinical feeding evaluations; however, there are few measures that are regularly used in children with CP or neurodevelopmental disability.[12] This impacts on clinicians' and researchers' ability to differentiate those with OPD from those with typical oral sensorimotor skills (discriminative assessment), to develop and evaluate more specific oral sensorimotor and nutritional interventions (evaluative assessment), and to predict outcomes based on the child's current level of oral sensorimotor function (predictive assessment).[13–15] There have been three reviews documenting measures of OPD in the neonatal age group.[6,12,16] Other authors have explored assessment of OPD in children with CP;[13,17] however, these reviews were not systematic. Hence, our aim was to conduct a systematic review investigating OPD measures for preschool children with CP or neurodevelopmental disabilities.

METHOD

Search strategy

A systematic search was conducted using computerized databases, including CINAHL (EBSCO), MEDLINE (1948 to February 2012), Cochrane Library (1970 to February 2012), AMED (1985 to February 2012), and PsycINFO (1987 to February 2012). The keywords used were ('oral motor' OR oromotor OR dysphagia OR feeding OR deglutition OR masticatory muscles) AND 'cerebral palsy' AND (child* OR p?ediatric) AND (assessment OR 'outcome measure' OR evaluat* OR psychometrics OR validity OR reliability) and corresponding Medical Subject Headings terms. A secondary search to include neurodevelopmental disabilities was conducted ('neurological disorder' OR 'physical impairment' OR 'movement disorder' OR disab*) owing to a lack of measures identified that were specific to CP.

Inclusion/exclusion criteria

Papers were included if they described measures that met the following criteria: (1) objective measures of OPD, defined as direct clinical assessment using standardized administration and scoring guidelines; (2) 40% of items related to ingestion functions on the ICF;[5] (3) evaluated published psychometric properties in children with CP[18] or neurodevelopmental disorders[19] aged 12 months to 5 years; and (4) were available for use in Australia.

Papers were excluded if they (1) did not measure OPD during the oral or pharyngeal phase associated with eating, drinking, or controlling saliva; (2) were unable to quantify OPD (including informal or non-standardized measures); (3) described a behavioural feeding assessment[20] rather than OPD; and (4) were not published in English (owing to a lack of translation services).

What this paper adds

- This is the first systematic review of objective measures of OPD in children with CP or neurodevelopmental disabilities aged 12 months to 5 years.
- The article summarizes the clinical utility, validity, and reliability of oropharyngeal dysphagia measures for this group of children.
- The Schedule for Oral Motor Assessment, the Functional Feeding Assessment, modified, and the Dysphagia Disorders Survey are identified as the strongest available measures for OPD.

Data extraction and quality assessment

Titles and abstracts were screened by two authors independently (KAB, KAW), and disagreements were discussed until a consensus was reached. Papers were retained if the means of assessing OPD were not identified in the title or abstract. Full-text copies of papers, administration manuals, and scoring sheets were sought for analysis of measure properties. These were evaluated against the inclusion criteria by two authors (KAB, KAW) to generate the final list of measures to be analysed.

Content analysis was completed using the ICF linking rules to determine measures meeting the 40% ingestion function inclusion criterion, with only the items that contributed to the total score counted.[21] The statistical strength of the psychometric properties of measures and the methodological quality of studies were evaluated using the scoring system of the Consensus-based Standards for the Selection of Measurement Instruments (COSMIN) Checklist[22,23] for which content validity and interrater reliability have been established.[23,24] An adapted version of the *CanChild* Outcome Rating Form was used to collect information on the measure characteristics (primary purpose of the measure, target population, structure, and domains tested) and clinical utility (procedures, time, manual/equipment, and qualifications/training). This was scored to provide an objective rating (see Appendix S1, supporting material online, for characteristics reviewed, definitions, and scoring).

The COSMIN taxonomy of measurement has been well defined.[25] The properties included in this review, definitions, and scoring guidelines are reported in Appendix SI. The statistical strength of each psychometric property was rated as good (+), intermediate (0), poor (–), or unknown (?) according to the COSMIN guidelines.[26] The methodological quality of studies reporting psychometric properties was rated as excellent, good, fair, or poor.[22] All studies with psychometric data available were included in this review, including those for which the primary purpose of the study was not to evaluate the measure's psychometrics. Methodological quality ratings were conducted only on the methods related to assessing the psychometric properties of OPD measures, and do not reflect the overall quality of the study. The 'worst score counts' principle[22] was used to give an aggregated methodological quality rating, combining all items except sample size, as a

small sample size affects the precision of the estimate rather than representing an overall methodological flaw. A confidence band was instead calculated for the 0.7 statistical cut point, which indicated that studies with a sample size or more than 20 were adequate for 95% confidence that the statistic is better than 'fair' (>0.3).

RESULTS

The systematic search identified 820 references, of which 62 full-text papers were sought after screening of the title and abstract. Review by two independent raters (KAB, KAW) identified 27 papers and nine measures that met the inclusion criteria. Measures included the Brief Assessment of Motor Function – Oral Motor Deglutition Scale (BAMF-OMD),[27] the Behavioral Assessment Scale of Oral Functions in Feeding,[28] the Dysphagia Disorders Survey (DDS),[29] the Feeding Behaviour Scale (FBS),[30] the Functional Feeding Assessment, modified (FFAm),[31] the Gisel Video Assessment (GVA),[32] the Oral Motor Assessment Scale (OMAS),[33] the Pre-Speech Assessment Scale (PSAS),[34] and the Schedule for Oral Motor Assessment (SOMA).[35] The process used to identify OPD measures and the results are shown in Figure 1. Appendix SI reports the excluded measures and reasons for exclusion.

The nine measures varied in their purpose, structure, and scoring system (Table I). Five measures were developed specifically to assess children with CP[30,32–35] and the other four for children with neurodevelopmental disabilities.[27–29,31] The primary purpose of six of the measures was evaluative,[27–29,31,32,34] and discriminative for the remaining three.[30,33,35] Four of the measures assessed the child's oral skills in everyday feeding (oral motor performance),[27,28,30,33] while five included test items aimed to assess a number of feeding behaviours across a range of food/fluid textures and utensils (oral motor capacity). All but two measures covered the major textures of purées, chewable solids, and fluids, and the other two were restricted to solids only (FBS and GVA). The inclusion of tough chewable foods, items examining different fluid utensils (cup, trainer cup, or cup and straw), saliva control, and a specific focus on the pharyngeal phase varied between measures. The total number of items ranged from as few as seven behaviours (OMAS) up to 80 on the SOMA and 89 on the FFAm.

The overall score for each measure's psychometric properties ranged from two on the FBS to 11 on the SOMA and the FFAm, out of a possible score of 24 (Table II). Details of each measure's validity are reported in Table II. The content validity of all measures was poor except for the PSAS and SOMA, whose contented validity was rated as moderate and strong respectively. The DDS showed statistically good convergent validity, with moderate methodological rigour, when compared with blinded speech pathologist assessment. All other studies showed uncertainty in the statistical strength and limited methodology for convergent validity. Three of five measures evaluated for discriminative validity showed a significant difference between cases and controls (FFAm, OMAS, SOMA). Responsiveness was evaluated only in the FFAm and GVA, and this was not statistically or methodologically strong.

Details of the reproducibility of measures are reported in Table III. Most measures showed statistically good reliability, although the methodological quality of the studies varied and the study sample sizes tended to be small. The FBS had no reliability tested. Test–retest reliability was strong in the BASOFF and the SOMA, moderate in the OMAS, and limited in the FFAm. For the SOMA, test–retest reliability was evaluated between bolus trials in the same mealtime rather than showing stability across time. Intrarater reliability was reported only for the BAMF-OMD and showed a high correlation between scores, but the study methodology was poor. Interrater reliability was assessed for all measures (except the FBS), generally with good statistical strength but with varying methodological quality. Measurement error could be calculated only for the FFAm. The change in scores not attributable to measurement error (smallest detectable change) was less than the minimum important change of 20%, indicating that a clinically meaningful change in scores is likely to be real.

The clinical utility of measures is detailed in Table IV. The DDS and SOMA scored a maximum score of nine for clinical utility. Five measures were observations of a typical mealtime, including a range of textures, and needed minimal child compliance. The FFAm, GVA, PSAS, and SOMA all required greater child compliance, as one or all of (1) textures, (2) procedures, and (3) utensils were standardized. The BAMF-OMD was the only measure that included parent report to gather information, and could also be completed face to face or from video. The FFAm, GVA, PSAS, and SOMA allowed video ratings of behaviours. The majority of the measures were completed during mealtime and required between 15 and 45 minutes, but administration time ranged from 5 minutes[27] up to 2.5 to 3 hours.[34] Only the DDS, PSAS, and SOMA had published manuals, with the DDS manual only available after certification training. The PSAS manual is available online, but has been superseded by a simplified checklist, which is available in the author's textbook.[38] Other measures reported administration and scoring information in journal articles only.

DISCUSSION

This systematic review identified nine objective measures of OPD that have published psychometric data in children with CP or neurodevelopmental disabilities aged 12 months to 5 years. Measures of OPD represent an important component of a comprehensive oral sensorimotor and mealtime assessment. There are few OPD measures available to clinicians that meet the informational needs for decision-making and have strong enough psychometric properties to provide confidence in the results. Owing to this paucity of measures for assessing OPD in preschool children with CP, there was no restriction in this review of inclusion of measures based on their purpose or domains, and the review was extended to include neurodevelopmental disabilities. This meant that the nine measures included in the review all varied in the type of information gathered and their practical application.

The comprehensiveness of measures varied in the range of food/fluid textures assessed, and the number and types of oral/pharyngeal behaviours included. This is important for

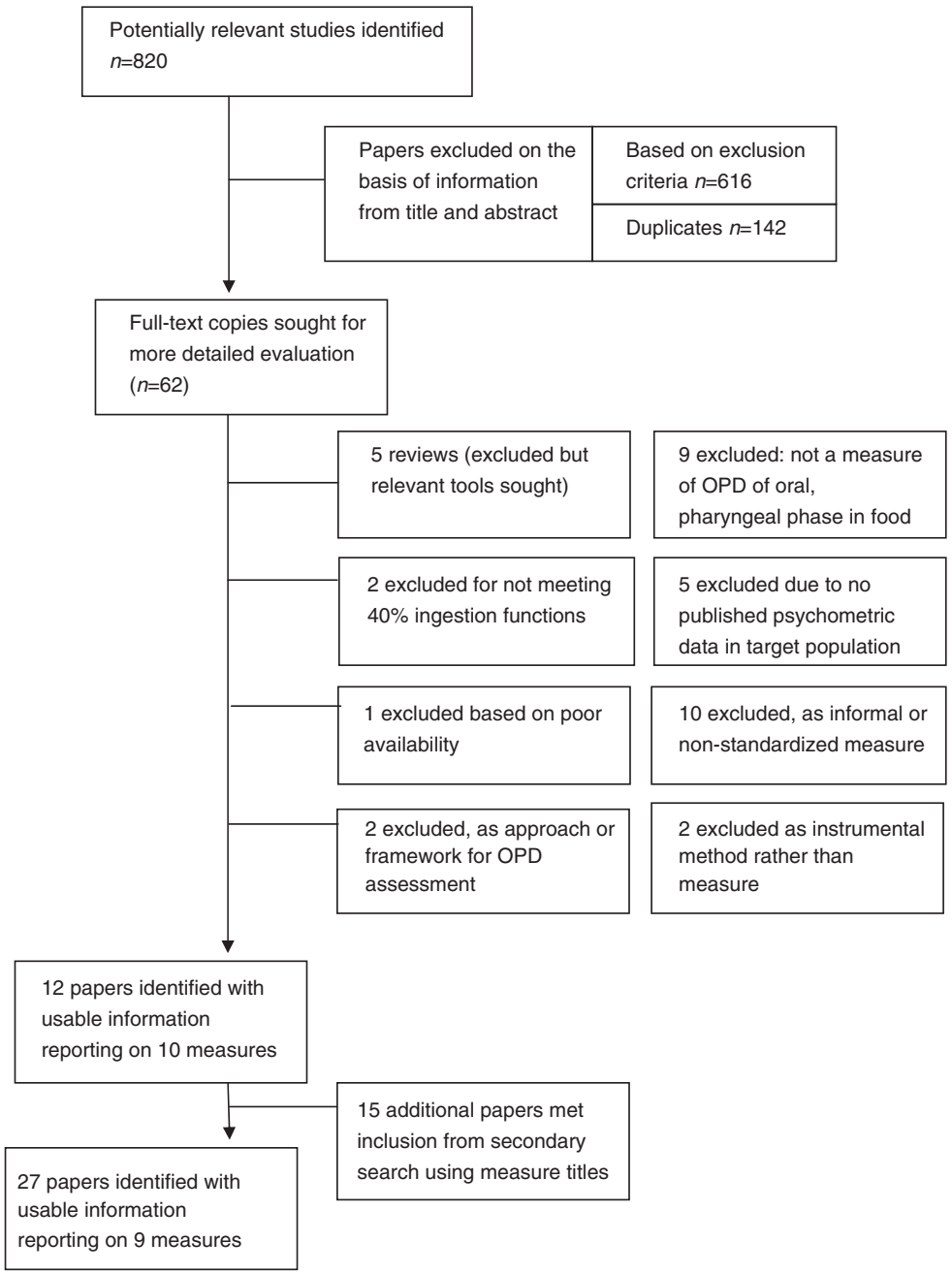

Figure 1: Processes performed to identify measures of oropharyngeal dysphagia and related papers. OPD, Oropharyngeal dysphagia.

clinicians and researchers to consider when deciding on a measure that is aligned with the purpose of the assessment. There was a comparable range of feeding domains included in all of the measures, but the number of items on each of these domains differed markedly. Many measures had 100% of items relating to ingestion functions, but this did not necessarily reflect the detail of analysis of individual oral/pharyngeal behaviours provided by the measure.

Overall, the validity of OPD measures was limited, with the highest validity score being 5 out of a possible score of 12 (for the SOMA). This is in part because the construct of OPD in this population remains poorly defined. There is no widely used theoretical foundation to describe OPD in children with CP, and no criterion standard or universally agreed definition of the particular parameters and performance encompassed by the term. Test construction has largely relied on literature review and expert opinion to determine item inclusion, combined inconsistently with a small pilot study to exclude or reword items with poor reliability. The purpose of the measure and the target populations were often not clearly defined. Without clearly defining these, it is difficult to assess the relevance of all items to the measure's purpose, population, or

Table I: Oropharyngeal measures for preschool children with cerebral palsy (CP) or neurodevelopmental disabilities: purpose and structure

Measure	Target population	Primary purpose	Performance or capacity	OPD domains[a]	Total items	% Ingest Func.	Scoring	Interpretation of scores
BAMF-OMD	Neurodevelopmental disabilities aged 6mo–18y	Evaluative	Performance	Fluids (3) Straw (1) Purée/semi-solid (1) Bite/chew (2) Tough chew (1) Saliva (1)	11	100	11-level scale	Classification on scale (0–10)
BASOFF	Developmental delay aged 10–38mo	Evaluative	Performance	Cup (1) Purée (2) Bite/chew (1) Swallow (5)	14	100	Nine behaviours	Ordinal scaled items (0, passive, to 5, normal); total score=sum of item scores
DDS	Developmental disability aged 3–13y	Evaluative	Capacity (aided)	Fluids (7) Purée (7) Bite/chew (8) Swallow (Y)	29	65	Two parts: dysphagia-related factors and swallowing competency	Binary scored items Raw scores Centiles
FBS	CP aged 2–16y	Discriminative	Performance	Purée (14) Bite/chew (14)	20	60	Six feeder behaviours; 14 child behaviours	Total number of behaviours Cut scores: normal, marginal, inadequate
FFAm	Neurological disability including CP aged 2–18y	Evaluative	Capacity	Cup (14) Straw (12) Purée/s-solid (16) Bite/chew (24) Swallow (12) Saliva (1)	89	79	Six domains, each with normal and abnormal behaviour lists	Ordinal scaled items (1, poor, to 5, normal) % Competence score for each domain and total
GVA	Cerebral palsy aged 2–16y	Evaluative	Capacity	Purée (1) Bite/chew (1) Tough chew (1)	9	100	Three food textures: purée, soft solid, viscous Time (s), cycles, Time–cycle ratio	Comparison with age norms (with SD)
OMAS	Cerebral palsy aged 3–13y	Discriminative and evaluative	Performance	Cup (1) Straw (1) Purée/s-solid (1) Bite/chew (3)	7	100	Seven behaviours	Ordinal scale (0, passive, to 3, functional) Classification score=most frequent score
PSAS	CP and neurological impairments aged 3–13y	Evaluative	Capacity (unaided)	Bottle (1) Cup (1) Purée (1) Bite/chew (4) Swallow (4) Saliva (1)	27	44	27 Performance areas (164 sub-items not scored individually)	Normal and abnormal scales Age norms
SOMA	NOFT and CP aged 10–42mo	Discriminative	Capacity	Bottle (9) Trainer cup (14) Cup (9) Purée (9) Semi-solid (8) Bite/chew (22) Swallow (Y) Saliva (Y)	80	98	Seven oral motor challenge categories, 8–22 behaviours per category	Binary scored items Cut scores (±OMD)

Discriminative assessment – used to distinguish between individuals based on dimension of interest, such as those with oropharyngeal dysphagia; evaluative assessment – used to measure change in an individual; predictive assessment – used to identify individuals who have or will develop an outcome of interest; capacity – measures what the individual is able to do; performance – measures what the individual typically does. [a]Number in brackets denotes the number of items testing the domain. Y indicates this domain is tested, but the item co-occurs with another domain, such as food texture. OPD, oropharyngeal dysphagia; Ingest Func., ingestion functions on the International Classification of Function; BAMF-OMD, Brief Assessment of Motor Function – Oral Motor Deglutition; BASOFF, Behavioral Assessment Scale of Oral Functions in Feeding; DDS, Dysphagia Disorders Survey; FBS, Feeding Behaviour Scale; FFAm, Functional Feeding Assessment modified; GVA, Gisel Video Assessment; OMAS, Oral Motor Assessment Scale; PSAS, Pre-Speech Assessment Scale; SOMA, Schedule for Oral Motor Assessment; OMD, oral motor dysfunction; CP cerebral palsy; NOFT, non-organic failure to thrive.

Table II: Oropharyngeal measures for preschool children with cerebral palsy (CP) or neurodevelopmental disabilities: validity

Measure	Overall psychometric rating	Content validity	Construct validity: convergent validity	Construct validity: discriminative validity	Responsiveness
BAMF-OMD	3	Limited (one poor study)[27] Expert panel (*n*=28) formally evaluated instrument against objectives. All items met a priori levels for agreement	Nil	Nil	Nil
BASOFF	6	Limited (one poor study)[28] Use in author's facility for over 3y with varying evaluators	Nil	Nil	Nil
DDS	4	Limited (one fair study)[29] Preliminary version validated *n*=626 participants aged 3–78y with DD Items with poor internal consistency/inter-item reliability deleted Pearson correlation coefficients to examine test structure ($p<0.05$)	Moderate (2 fair studies) **(+)**, *r*=0.84–0.97, *p*=0.0005 **(+)**[29;] Comparison with SP diagnosis, *n*=626 participants aged 3–78y with DD; *r*=0.92 **(+)**[43;] Comparison with SP diagnosis *n*=427 (mean age 33y)	Nil	Nil
FBS	2	Nil	Limited (two poor studies) (?), *r*=0.072, *p*=0.878 (–)[30.a;] Comparison with GVA (feeding efficiency), *n*=7 participants aged 2–16y with CP; Highest behaviour score clinically judged to have most severe feeding problems (?)[30,a;] Comparison with clinical judgement (whose judgement not identified), *n*=7 participants aged 2–16y with CP	Limited (one poor study)[30,a] No statistics reported (scores all marginal or abnormal for children with CP; no mention of comparisons)	Nil
FFAm	11	MFP: limited (one fair study)[41] Preliminary version: 198 items developed by nine experts Validated on *n*=8 (age not defined) Items with poor interrater reliability reworded or discarded	Limited (one fair, three poor studies) (?), *r*=0.54, $p<0.001$ (0)[30,a;] Comparison with GVA (feeding efficiency), Sample not described, *r*=0.61, $p<0.001$ **(0)** (OPT),[44] *r*=0.72, $p<0.001$ **(+)** (OFMF); Comparison with OPT/OFMF (oral praxis), *n*=48 aged 4–16y, mild CP, *r*=0–0.62 **(0)**[44;] Comparison to GVA (feeding efficiency), *n*=48 aged 4–16y, mild CP, *r*=–0.88, *p*=0.02 (+)[36,a] Comparison with drooling severity, *n*=6 children aged 4–13y with CP	Limited (one fair study) (+) $p<0.001$ (+)[44] Spoon feeding, biting, cup drinking, and swallowing differed significantly between participants and comparison children 87–98% competence for children with CP, compared with 100% competence for comparison children on all but two subtests (98–99%) *n*=48 children aged 4–16y with mild CP and a comparison group	Limited (one poor study) (–) Mean improvements from 10–19% during treatment (–)[36,a] Statistically significant improvements for 4/6 domains *n*=6 aged 4–13y with CP drooling and dysphagia

Table II: Continued

Measure	Overall psychometric rating	Content validity	Construct validity: convergent validity	Construct validity: discriminative validity	Responsiveness
GVA	6	Nil	Limited (four poor studies) (?) r=0.072, p=0.878 (–)[30,a;] Comparison to FBS, n=7 children aged 2–16y with CP, r=0.5–0.54, p<0.001–0.003 **(+)** for hard solid and purée)[44;] Comparison with FFAm n=48 children aged 4–16y with mild CP, r=03.44–0.46, p<0.001 (± for hard solid and purée)[44;] Comparison with OFMF (oral praxis), n=48 children aged 4–16y with mild CP, r=0.54, p<0.001 (+)[30,a;] Comparison with FFAm, sample not described	Limited (one fair study, one poor study) (?) p<0.001 for hard solid, but no significance for other textures (?)[44] n=48 aged 4–16y, mild CP p<0.001 for purée⁄s[30,a] p<0.001 for 4⁄7 children for solids (?) n=7 children aged 2–16y with CP	Limited (one poor study) F=17.53, p=0.006, for purée(s)[45,a] F=7.17, p=0.044, for viscose solid (increased time) (+) n=7 children aged 4–16y with CP and aspiration
OMAS	7	Limited (one poor study)[33] Constructed according to theory of measurement process; Conceptual framework through review of concepts of motor oral function; Items elaborated by MDT	Nil	Moderate (one good study) **(+)** Mann–Whitney U test (p<0.001) **(+)**[33;] n=53 children aged 3–13y, CP; n=54 age-matched comparison children	Nil
PSAS	3	Moderate (one good study)[34] Literature review and clinical experience to develop items Seven revisions including field testing for 8y by 215 trained clinicians (yearly feedback) – over 800 scored assessments	Nil	Nil	Nil
SOMA	11	Strong (one excellent study)[39] Literature review and clinical experience to develop items 75–90 items per domain Items with poor interrater reliability or high refusal rate were excluded	Limited (two poor studies) (?), r=0.76–0.82 (+)[46,a;] Resolution of swallowing impairment on PHAD and SOMA: week 8 all impairment resolved (SOMA), 15% mild impairment (PHAD), n=13 children aged 4–15y with TBI 20% had OMD on SOMA, 84% had dietary modifications **(?)**[40,a;] n=28 children aged 2;6–16;5y with Worster–Drought syndrome	Limited (one fair study) **(+)** F=42.43, p<0.001 **(+)** for overall test[47;] 10–80% false negatives for CP for individual OMCCs (no statistics); n=127 children aged 8–42mo (58 comparison children, 56 NOFT, 12 CP)	Nil

Methodological quality rated as limited, moderate, or strong. Statistical strength identified in brackets as +, good; 0, intermediate; –, poor; ?, unknown. Statistic rating bold if the study sample was over 20, indicating adequate sample size for 95% confidence that statistic is better than fair (0.3). [a]Primary purpose of paper was not to evaluate measure psychometrics; content validity – content of the tool measures the construct intended; convergent validity – tests relationship between a new assessment and existing valid tool; discriminative validity – ability to differentiate between two constructs; responsiveness – ability to detect change over time (for full definitions see Appendix SI). BAMF-OMD, Brief Assessment of Motor Function – Oral Motor Deglutition; BASOFF, Behavioral Assessment Scale of Oral Functions in Feeding; DDS, Dysphagia Disorders Survey; DD, developmental disability; SP, speech pathologist; FBS, Feeding Behaviour Scale; GVA, Gisel Video Assessment; FFAm, Functional Feeding Assessment, modified; MFP, Multidisciplinary Feeding Profile; OPT, Oral Praxis Test; OFMF, oral-facial motor function; OMAS, Oral Motor Assessment Scale; SOMA, Schedule for Oral Motor Assessment; PHAD, Paramatta Hospitals Assessment of Dysphagia; TBI, traumatic brain injury; PSAS, Pre-Speech Assessment Scale; OMCC, Oral Motor Challenge Category (subtest on SOMA); OMD, oral motor dysfunction; MDT, multidisciplinary team; NOFT, non-organic failure to thrive.

Table III: Oropharyngeal measures for preschool children with cerebral palsy (CP) or neurodevelopmental disabilities: reliability

Measure	Test–retest reliability	Intrarater reliability	Interrater reliability	Measurement error
BAMF-OMD	Nil	Limited (one poor study) **(+)**[27] r=0.997, p<0.001 **(+);** n=20 children aged 6mo–18y with DD (n=18) and a comparison group (n=2)	Limited (one poor study) **(+)**[27] Kendall's W=0.977, p<0.001 **(+);** n=20 children aged 6mo–18y with DD (n=18) and comparison children (n=2)	Nil
BASOFF	Strong (one excellent study) **(+)**[48;] ICC=0.68–0.79 **(+),** n=46 children aged 1–21y with DD	Nil	Limited (two studies: one good, one poor) **(+),** ICC=0.72–0.76 **(+)**[48;] n=46 participants aged 1–21y with DD; ICC=0.96 (+)[49,a;] n=3 children aged 3–15y with severe ABI	Nil
DDS	Nil	Nil	Limited (one poor study) **(+)**[29;] 97% agreement (following discussion) **(+),** n=21 (sample characteristics not reported)	Nil
FBS	Nil	Nil	Nil	Nil
FFAm	Limited (one poor study) (+)[36,α] Change in means scores not significant (p>0.05) (+) n=15 children aged 4–13y with CP	Nil	Strong (two good studies) **(+);** ICC=0.89 (for CP group alone ICC=0.93) **(+)**[44;] n=48 children aged 4–16y with mild CP or comparisons Concordance correlation=0.823–0.984 (+)[36, a;] n=15 children aged 4–13y with CP	Moderate (one good study) **(+);** Change during control –2.8% to +4.3%[36, a] LoA smaller than 20% (MIC) for cup, straw, swallow SDC 6.1–14.7% n=15 children aged 4–13y with CP
GVA	Nil	Nil	Strong (five good studies) (0/+); ICC=0.67–0.99 (0/+)[50,a;] n=18 children aged 2–4y or comparison children; ICC=0.88–0.97 **(+)**[32, a;] n=98 children aged 5–8y or comparison children Independent t-test, p=0.802–0.069; **(+)**[51,a;] n=143 children aged 6mo–2y, controls; ICC=0.69–0.97 (lower for purée/s) **(0/+)**[44;] n=48 children aged 4–16y with mild CP or comparison children ICC=0.85–0.95 **(+)**[45,a;] n=28 aged 3–8y with CP	Nil
OMAS	Moderate (one good study) **(+);** κ=0.872–1.0 for items and κ=1.0 overall **(+)**[33;] n=53 children aged 3–13y with CP	Nil	Moderate (one good study) **(+);** κ=0.892–1.0 for items and κ=1.0 overall **(+)**[33;] n=53 children aged 3–13y with CP	Nil
PSAS	Nil	Nil	Limited (one poor study) **(0/+);** 65–87% agreement with redetermined standard of correctness **(0/+)**[34;] n=78 (sample characteristics not specified)	Nil
SOMA	Strong (one excellent study, one fair) **(+);** κ=1.0 for 84% **(+)**[39] Comparison of trials 1 and 3 n=10 children aged 8–24mo (seven with NOFT, three comparison children); κ=1.0 for 78% (0)[52, a;] n=6 aged 11–27mo with DS (and twin comparisons)	Nil	Strong (one excellent study, one fair) (0); κ=1.0 for 68% (0)[39] n=10 (three trials per child) children aged 8–24mo (seven with NOFT, three comparison children) Reliability for CP, no statistics 'similar findings to above trial'; κ=1.0 for 56% (0)[52,a;] n=6 children aged 11–27mo with DS (and twin comparisons)	Nil

Methodological quality rated as limited, moderate, or strong. Statistical strength identified in brackets as: +, good; 0, intermediate; –, poor; ?, unknown. Statistic rating bold if the study sample was more than 20, indicating adequate sample size for 95% confidence that statistic is better than fair (0.3). [a]Primary purpose of paper was not to evaluate measure psychometrics; test–retest reliability – consistency on repeated measures over time; intrarater reliability – consistency on repeated measures by same rater on different occasions; interrater reliability – consistency on repeated measures by different raters; measurement error – error in score unrelated to true changes (for full definitions see Appendix SI). BAMF-OMD, Brief Assessment of Motor Function – Oral Motor Deglutition; DD, developmental disability; BASOFF, Behavioral Assessment Scale of Oral Functions in Feeding; ICC, intraclass correlation coefficient; DDS, Dysphagia Disorders Survey; FBS, Feeding Behaviour Scale; FFAm, Functional Feeding Assessment, modified; LoA, limits of agreement; MIC, minimal important change; SDC, smallest detectable change; GVA, Gisel Video Assessment; OMAS, Oral Motor Assessment Scale; PSAS, Pre-Speech Assessment Scale; SOMA, Schedule for Oral Motor Assessment; DS, Down syndrome; NOFT, non-organic failure to thrive.

Table IV: Oropharyngeal measures for preschool children with cerebral palsy or neurodevelopmental disabilities: clinical utility

Measure	Overall rating (max 9)	Perspective	Procedure and (child compliance)	Rating	Administration time	Manual/ equipment	Clarity	Items and (details)	Availability and qualifications
BAMF-OMD	7	Professional (and parent interview)	Observational (low)	Video, face to face and parent report	<5min	No manual: form in journal[27] (1)	3	3 (Brief)	No qualifications/training specified
BASOFF	5	Therapists	Observational (low)	Face to face	Not specified – 20–30min	No manual: form in journal[28] (1)	3	1 (Brief)	No qualifications/training specified
DDS	9	Professional	Observational (low)	Face to face	Not specified – 30–45min	Manual available after training (US$300) (3)	3	3 (Med)	Certification training. Run on a request basis in Australia
FBS	3	Professional	Observational (low)	Face to face	20–30min	No manual: brief description in journal[30] (1)	1	1 (Brief)	No qualifications specified
GVA	5	Professional	Standardized textures (medium)	Video		Norm charts in Schwaab et al.[50] (1)	2	2 (Brief)	No qualifications specified
FFAm	6	Feeding professionals	Some standardized components (medium)	Face to face or video	30–45min	Draught manual developed, but not available: form in Kenny et al.[41] and modification guidelines in Gisel[31] (1)	2	3 (Med)	No qualifications/training specified
OMAS	5	Feeding professional	Observational (low)	Face to face	Not specified – 15min	No manual: form in journal[33] (1)	1	3 (Brief)	No qualifications/training specified
PSAS	6	Feeding professional	Observational with required textures (medium)	Face to face or video	2.5–3h + complex scoring	1982 manual available online. Checklist version available in textbook (US$89.99) (2)	1	3 (Long)	Originally 4d training course (no longer running). To use, examiner must spend significant time practising administration and scoring
SOMA	9	Allied health	Standardized textures and administration (medium–high)	Video	15–20min	Manual (US$198) (3)	3	3 (Medium)	Assessment not currently available (2011)

BAMF-OMD Brief Assessment of Motor Function – Oral Motor Deglutition; BASOFF, Behavioral Assessment Scale of Oral Functions in Feeding; DDS, Dysphagia Disorders Survey; FBS, Feeding Behaviour Scale; GVA, Gisel Video Assessment; FFAm, Functional Feeding Assessment, modified; OMAS, Oral Motor Assessment Scale; PSAS, Pre-Speech Assessment Scale; SOMA, Schedule for Oral Motor Assessment.

construct, which are all part of the rigorous content validation methodology. The SOMA, a discriminative assessment, was the only measure that reported on excluding items that failed to provide reasonable discrimination between groups. The PSAS construction was based on a neurodevelopmental theory, although items were not assessed to determine whether the construct was comprehensively reflected. The purpose of many of the measures was evaluative (i.e. to measure change in individuals), although all but two measures did not investigate the ability to detect change (responsiveness) after treatment. The responsiveness of the FFAm and GVA was investigated in a single study with limited methodology. Children's score change on the FFAm after treatment was less than the clinically important change of 20%,[36] and the GVA showed significant change for purées only. Without knowing a measure's ability to detect clinically important change (i.e. the measure's responsiveness), its application as an evaluative measure in intervention studies and clinical practice is limited.

The reliability of most measures was good, although this needs to be confirmed with studies of higher methodological rigour and larger sample size. Ingestion functions can be difficult to consistently observe, as it can be hard to visualize movements occurring inside the oral cavity or pharynx. A number of measures used the reliability of items during their construction to exclude items with poor reliability, which meant that the retained items were those that could be more reliably observed. This is reflected in the good statistical results of many of the reliability studies. The exclusion of items with poor reliability during measure construction may result in oral/pharyngeal behaviours that are clinically important or responsive to treatment but are not being measured, which presents a limitation of the measures of OPD. Interrater reliability was studied across all but one measure, and all other types of reliability were inconsistently explored.

Mealtime assessment can be challenging, as a functional assessment includes motor, sensory, behavioural, structural, and personal factors, such as past experiences and family preferences.[37] Scores may be significantly influenced by performance during a particular mealtime. There has been limited exploration of repeated administration of measures (test–retest reliability), but some authors have attempted to control some of the variability arising from individual mealtime factors by standardizing the foods, utensils, position, and presentation of assessments. Although it has a positive impact on the reproducibility of assessments, allowing a more naturalistic mealtime permits the clinician to evaluate the child's typical performance. Understanding the test–retest reliability of measures across time is imperative if they are to be used longitudinally in research or clinical contexts.

The clinical utility of OPD measures was limited mostly by their lack of published manuals. Only the DDS, PSAS, and SOMA had manuals providing sufficient detail to ensure consistency in their use. The PSAS has been preferred by clinicians because of its comprehensiveness, but it takes a significant amount of time to administer and score. The PSAS was superseded by a checklist version, which is available in the author's textbook, but this does not allow the quantification of OPD.[38] Only a scanned copy of the original typewriter version of the PSAS is available (online). All of the measures were administered during mealtime, which means that they are acceptable to the child and family, representing a strength of OPD measures. The level of child compliance needed for measures with greater standardization or that assess a greater number of behaviours, such as the FFAm, GVA, PSAS, and SOMA, may restrict their use in a younger age range (e.g. 18–36mo) or result in missing data due to refusals to comply. A relatively high rate of refusals was reported in two studies using the SOMA, ranging from 17 to 41%.[39,40] The only measure that requires users to undergo certification training was the DDS, which may preclude it from wider utilization owing to the costs and availability of the training.

There is no single measure that represents a systematic and comprehensive evaluation of OPD in feeding for the clinical setting. Brief measures such as the BAMF-OMD and OMAS may prove useful for service planning or evaluation of a child across facilities, but they do not provide detailed information for individual clinical assessment or intervention planning. The SOMA and DDS were the most comprehensive measures, with good clinical utility and sound psychometric properties. Clinicians need to be clear on the purpose of their assessment and select measures accordingly.

The FFAm and SOMA are the measures that showed the highest level of psychometric rigour, although additional studies are needed to provide greater confidence for use in research. The modification to the original FFA subtest on the Multidisciplinary Feeding Profile[41] has been subject to criticism, suggesting that altered weightings void the measure's validity.[42] The translation of the ordinal scores of each item to an overall percentage competence score may distort the relative contribution of items, which should be considered if using the FFAm. The descriptions for scoring the FFAm are adequate for achieving good reliability, but the lack of a comprehensive scoring and administration manual may limit its use. To date, only the SOMA has been shown to be valid and reliable for use as a complete measure to differentiate those with normal skills from clinically significant OPD, although its sensitivity and specificity in detecting this are not reported. Using the SOMA oral motor challenge categories (texture and utensil subtests) individually will not provide accurate findings, as the method of validation leaves significant gaps in the measure's sensitivity in detecting all cases at this level. In addition, it has not been evaluated whether this measure is stable across time or valid as an evaluative measure.

The theoretical construct of OPD lacks definition, and this is reflected in the measures available. Further studies are needed to delineate whether a distinction should be made between dysfunction and delay when considering OPD in young children, and whether skills in assessment represent reduced motor capacity (what a child is able to do in a controlled environment), reduced capability (what a child can do in their daily environment), or poor motor performance (what a child actually does in a given environment). Children's feeding skills vary with age, but most measures cover a broad age range and do not provide age norms. Large normative studies in the

preschool age range would be beneficial by providing clinicians with age norms on measures, as well as contributing to the discriminative validity. In addition, there is a lack of measures that have been shown to be stable over time and responsive to change, which are essential before findings from intervention studies can be considered with confidence. There is a need for more studies exploring the reliability and responsiveness of commonly used measures, and for more comprehensive construct validation in the absence of a criterion standard. Establishing predictive validity by following changes in OPD over time and establishing an association between scores and health outcomes of interest will improve the validity of measures and contribute to an improved definition of the construct of OPD.

CONCLUSION

Nine objective measures of OPD in young children with CP and neurodevelopmental disability were identified from the 27 papers assessed in this review. These measures are not readily compared because of the range of measure purposes and domains. There is a paucity of measures of OPD with demonstrated psychometric properties available for this population. The SOMA and FFAm had the strongest psychometric properties of validity and reliability, and were most suitable for use in a research context. The SOMA and DDS had the strongest clinical utility to support clinical decision-making. Further studies to test the psychometric properties of existing measures would improve their potential for use in both clinical and research contexts. In particular, there is a distinct need to determine whether measures are stable across time (test–retest) and are able to detect change (responsiveness), as well as their predictive validity; the determination of these will contribute to an improved definition of the construct of OPD.

ACKNOWLEDGEMENTS

This project was supported by the National Health and Medical Research Council Postgraduate Medical and Dental Scholarship (1018264 – KAB), Career Development Fellowship (1037220 – RNB), and NHMRC Research Grants (569605) and (465128).

SUPPORTING INFORMATION

Additional material and supporting information for this paper may be found online.

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Appendix A: COSMIN taxonomy and scoring guidelines

Psychometric Property	Definition	Statistical strength:	Methodological strength* (total score=24)
VALIDITY			
Content validity	The degree to which the content of the tool measures the construct intended.	+: A clear description provided of the measurement aim, target population, concepts being measured and item selection, including a pilot study	*Strong=3*: consistent findings in multiple studies of good methodological quality or 1 excellent study *Moderate=2*: consistent findings in multiple studies of fair methodological quality or in 1 good study *Limited=1*: 1 study of fair methodological quality *Conflicting=1* *Unknown=1*: only studies of poor methodological quality
Hypothesis testing (construct validity): convergent	Tests a pre-defined relationship between a new assessment and existing valid tool which measures the same or similar construct.	+: Results in agreement with hypotheses≥75%. If no hypotheses were reported: Correlation to other measure>0.7	
Hypothesis testing (construct validity): discriminative	The tool's ability to differentiate between two constructs, for example those with OMD and those without.	+: $p<0.05$ for the difference between cases and controls	
Responsiveness	The tool's ability to detect small but clinically important change in performance over time, and distinguish clinically important change from measurement error.	+: MIC>SDC or MIC outside LoA	
REPRODUCIBILITY			
Test-retest reliability	The ability of the tool to score consistently when used to assess stable individuals on repeated measures over time.	+: Reliability statistic ≥0.70. If per-item percentage reported: ≥80% perfect agreement.	
Intra-rater reliability	The ability of the tool to score consistently when used to assess stable individuals when administered by the same person on different occasions.		
Inter-rater reliability	The ability of the tool to score consistently when used to assess stable individuals on repeated measures administered by different people.		
Measurement error	Error in an individual's score unrelated to true changes in the construct measured.		

Key: + good; 0 intermediate; - poor; ? unknown; MIC Minimal Important Change: clinically important change in scores, SDC Smallest Detectable Change: reflects the smallest within-person change in score that can be interpreted as real change above measurement error (with $p<0.05$), LoA Limits of Agreement: equals the mean change in scores of repeated measurements ±1.96 x standard deviation of the change.

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Update to Oropharyngeal Measures

Since the publication of the systematic review in 2012, there have been some developments in the available measures of OPD. Of note, is the publication of a new classification system, the *Eating and Drinking Ability Classification System* (EDACS),[73] for which the candidate participated in the Delphi survey. The candidate includes this classification in this update to OPD measures, to highlight its potential promise as a consistent framework in future OPD research. However, it is important to note that this classification system did not meet inclusion into the systematic review (not a 'measure' of OPD).

i. The EDACS was published in early 2014 to complement the existing classification systems for individuals with CP: the GMFCS, *Manual Ability Classification Syste*m (MACS), and *Communication Function Classification System* (CFCS).[74] It aims to reliably classify children's eating and drinking ability on a I-V level scale, and to create consensus in the severity levels used both clinically and in the research literature. The EDACS was found to be a valid and reliable system for children with CP aged 4 to 22 years, with absolute agreement for 78% of ratings (κ=0.72) between 2 speech therapists and 58% of ratings (κ=0.45) between a speech therapist and parent.[73]

ii. Two new measures of OPD have been developed which met the inclusion criteria of the clinimetric review: the *Mastication Observation and Evaluation Instrument*,[75] and the *Ability for Basic Feeding and Swallowing Scale for Children*.[76] Both of these measures have had limited testing of their psychometric properties (as described) and therefore these additional measures do not change the conclusions of the published review.

    - The *Mastication Observation and Evaluation Instrument*[75] was developed through a Delphi Study in response to the lack of valid and reliable measures (citing the results of our systematic review). The authors comment that the SOMA and FFAm (the best performing measures with regards to psychometric properties) had insufficient detail for assessing mastication, were not available in Dutch, and training for clinicians was unavailable. The *Mastication Observation and Evaluation Instrument* has 8 items scored on an ordinal 4-point scale, but it is limited to chewable foods (bread and biscuit).[77] This measure had its psychometrics tested in 2 studies: the first in 10 children with TD aged 11 to 42 months, and 10 children with CP aged 29 to 65 months;[75] and the second in 80 children with TD aged 6 to 48 months, and 44 children with CP aged 24 to 72

months (GMFCS II-IV).[77] Reproducibility was considered strong across both studies; intrarater ICC=0.73-1.0;[75] and interrater ICC=0.68-1.0, interrater Gwet's AC_2=0.77 for bread and AC_2=0.81 for biscuit.[77]

- The *Ability for Basic Feeding and Swallowing Scale for Children*[76] was again developed in response to a lack of satisfactory assessment scales available for childhood dysphagia. The authors' requirements were for an assessment scale of childhood dysphagia which could be administered by a range of different disciplines and family members. Their scale consists of 5 items rated on an ordinal 4-point scale, including the items of wakefulness, head control, hypersensitivity, oral motor ability, and saliva control. The scale was validated on 54 children with dysphagia aged 2 months to 14 years (3 with CP). Correlation was tested against 2 Japanese scales used predominately in adults, the *Fujishima Grade of Feeding* (R=0.322, significant) *and Swallowing Ability* and *Food Intake LEVEL Scale* (not significant).[76] Two of the five items showed almost perfect reliability, 2 moderate and 1 poor, although no overall reliability was reported.

iii. A paper describing the development and validation of the DDS was published earlier this year by Dr Sheppard.[78] The results presented in this paper were a re-analysis of the original validity and reliability data described in the test manual.[66] These findings have, therefore, already been synthesised in the clinimetric review presented in this chapter.

Summary of Chapter 2

Our review identified 9 objective measures of OPD with published psychometric data in preschool children with CP or neurodevelopmental disabilities. The SOMA and *Functional Feeding Assessment modified* (FFAm) had the strongest psychometric properties, and the SOMA and DDS the strongest clinical utility. The following key findings contributed to the selection and interpretation of measures in the current study, as well providing information for future clinical and research use.

i. The psychometrics of many OPD measures have had limited testing or testing in low-quality studies (according to the *Consensus-Based Standards for the Selection of Health Measurement Instruments* [COSMIN]).[79] Owing to the paucity of measures, no restrictions were placed on the inclusion criteria with regards to the domains assessed (eg, the food/ fluid textures, or phases of the swallow) or primary purpose of the

assessment (eg, designed to detect OPD, or measure change in skills following maturation or intervention). All measures reviewed (except the *Gisel Video Assessment* [GVA][30] and *Feeding Behaviour Scale* [FBS][80]) covered a minimum skill set of ingestion functions needed to consume a puree, chewable food and fluid, however the number of functions assessed on each varied significantly.

ii. Most OPD measures had limited validity, with the strongest validity found for the SOMA, with a maximum score of 5 out of 12. The validity of measures was influenced by a poorly defined construct of OPD.

iii. Only the FFAm[81] and GVA had their responsiveness tested (ie, ability to detect change), and these were in low-quality studies, and showed limited change. Without measures with strong responsiveness, the evaluation of intervention studies will be limited.

iv. All measures (except the FBS) had their interrater reliability tested, and it was generally found to be good (although methodological rigour of studies was lacking). A number of authors used reliability in the measure construction, excluding those items with poor reliability. This resulted in more reliable measures, but may mean clinically important behaviours are not assessed.

v. Only the *Brief Assessment of Motor Function, Oral Motor Deglutition* scale (BAMF-OMD)[82] had its intrarater reliability tested.

vi. Test-retest reliability was explored in 4 measures. It was found to be strong for the *Behavioural Assessment Scale of Oral Functions in Feeding*[83] and SOMA, although the SOMA only measured reliability between boluses rather than between mealtimes. Test-retest reproducibility tested between mealtimes is an important property for the longitudinal aspect of the doctoral research.

vii. The DDS and SOMA had the best clinical utility, as they had clear published manuals and provided detailed information about the child's performance. The PSAS, while scoring lower, was considered to provide the most comprehensive and clinically useful information, although the out-dated manual deemed hard to follow.

Having identified the measures with the strongest psychometrics and clinical utility for assessing OPD in preschool children with CP, this doctoral program aimed to determine the prevalence and patterns of OPD in this subgroup of children. The following chapter describes the protocol for the proposed doctoral reserach.

Chapter 3: Study Protocol

Introduction to Chapter 3

This chapter provides an introduction to substudies 1 to 3 which relate directly to the larger *Queensland CP Child: Growth, Nutrition and Physical Activity Study* (GNPA, NHMRC 569605);[84] and concurrent study, *Queensland CP Child: Motor Development and Brain Function Study* (NHMRC 465128).[85] It includes the published protocol paper, "Longitudinal Cohort Protocol Study of Oropharyngeal Dysphagia: Relationships to Gross Motor Attainment, Growth and Nutritional Status in Preschool Children with Cerebral Palsy" which describes the OPD literature (until May 2012) and methods of these substudies. The protocol paper is supplemented in this chapter with new papers from a current literature search (11 May 2012 to 15 December 2014).

Paper 2: Protocol Paper: Longitudinal Cohort Study of Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy

Benfer KA, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Longitudinal cohort protocol study of oropharyngeal dysphagia: relationships to gross motor attainment, growth and nutritional status in preschool children with cerebral palsy. *BMJ Open* 2012;2(4):e001460. http://bmjopen.bmj.com/content/2/4/e001460.full.pdf. Accessed August 27, 2012.

BMJ open accessible medical research

Longitudinal cohort protocol study of oropharyngeal dysphagia: relationships to gross motor attainment, growth and nutritional status in preschool children with cerebral palsy

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To cite: Benfer KA, Weir KA, Bell KL, *et al*. Longitudinal cohort protocol study of oropharyngeal dysphagia: relationships to gross motor attainment, growth and nutritional status in preschool children with cerebral palsy. *BMJ Open* 2012;**0**:e001460. doi:10.1136/bmjopen-2012-001460

▸ Prepublication history and additional material for this paper are available online. To view these files please visit the journal online (http://dx.doi.org/10.1136/bmjopen-2012-001460).

Received 11 May 2012
Accepted 29 June 2012

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ABSTRACT

Introduction: The prevalence of oropharyngeal dysphagia (OPD) in children with cerebral palsy (CP) is estimated to be between 19% and 99%. OPD can impact on children's growth, nutrition and overall health. Despite the growing recognition of the extent and significance of health issues relating to OPD in children with CP, lack of knowledge of its profile in this subpopulation remains. This study aims to investigate the relationship between OPD, attainment of gross motor skills, growth and nutritional status in young children with CP at and between two crucial age points, 18–24 and 36 months, corrected age.

Methods and analysis: This prospective longitudinal population-based study aims to recruit a total of 200 children with CP born in Queensland, Australia between 1 September 2006 and 31 December 2009 (60 per birth-year). Outcomes include clinically assessed OPD (Schedule for Oral Motor Assessment, Dysphagia Disorders Survey, Pre-Speech Assessment Scale, signs suggestive of pharyngeal phase impairment, Thomas-Stonell and Greenberg Saliva Severity Scale), parent-reported OPD on a feeding questionnaire, gross motor skills (Gross Motor Function Measure, Gross Motor Function Classification System and motor type), growth and nutritional status (linear growth and body composition) and dietary intake (3 day food record). The strength of relationship between outcome and exposure variables will be analysed using regression modelling with ORs and relative risk ratios.

Ethics and dissemination: This protocol describes a study that provides the first large population-based study of OPD in a representative sample of preschool children with CP, using direct clinical assessment. Ethics has been obtained through the University of Queensland Medical Research Ethics Committee, the Children's Health Services District Ethics Committee, and at other regional and organisational ethics committees. Results are planned to be disseminated in six papers submitted to peer reviewed journals, and presentations at relevant international conferences.

INTRODUCTION

Children with cerebral palsy (CP) may have poor feeding skills, influencing their growth, nutrition and overall health.[1 2] CP is the most common cause of physical disability in childhood, estimated at 2 per 1000 live born infants within Australia.[3] CP is an umbrella term which describes a group of disorders of movement and/or posture and motor function, which is permanent but not unchanging and due to a non-progressive interference/lesion in the developing brain.[4] Individuals with CP are a heterogeneous group, varying by severity and extent of motor involvement, type of movement patterns, aetiology and related conditions.[3]

The neurological lesion associated with CP may impact on the muscles of the jaw, cheeks, lips, tongue, palate and pharynx,[5] which manifest functionally as difficulties with controlling saliva, eating, drinking, swallowing and speaking. Eating and drinking are complex sensorimotor activities, which can be described in four phases, including the oral-preparatory, oral (propulsive), pharyngeal and oesophageal phases of the swallow.[6] This study will focus on oropharyngeal dysphagia (OPD) in young children with CP, defined as impairment to any component of the oral and/or pharyngeal phases associated with eating, drinking or controlling saliva.

The oral-preparatory phase is initiated when food/fluid is taken into the mouth, and involves tasks necessary in bolus formation, including sucking, munching and chewing. Food and fluid are contained in the oral cavity surrounded by the upper dental arch and closure of the lips. Posterior leakage of the fluid bolus is prevented by contact between the soft palate and tongue;

however, this contact is not maintained during the processing of the solid food bolus. The oral (propulsive) phase involves the backward propulsion of the food bolus, by the tongue gradually expanding its contact with the hard palate posteriorly, to initiate the pharyngeal swallow.[6 7] The duration and movements necessary for the oral phases differ depending on the child's age and the utensils used to ingest food/fluid.[7] The oral-preparatory phase of the swallow also differs when ingesting food compared to fluid boluses. When defining the swallow stages for solid foods, Matsuo and Palmer[6] advocate the use of the Process Model of Feeding, because of the overlap between the phases described in the Four Stage/Phase Model for fluids. The Process Model divides the oral-preparatory phase into Stage I Transport and Food Processing, in which the food is first ingested and moved onto the lateral occlusal surfaces of the teeth before being masticated to an optimal consistency for swallowing.

The pharyngeal phase is used to describe the passage of both food and fluid boluses through the pharynx, although when ingesting fluids it normally overlaps with the oral propulsive phase.[6] On initiation of the pharyngeal phase, the soft palate elevates to seal the nasopharynx to prevent nasal regurgitation. The tongue base retracts, propelling the bolus posteriorly against the pharyngeal walls followed by the pharyngeal constrictor muscles contracting to squeeze the bolus downward. To ensure airway safety during bolus passage, respiration ceases momentarily, the vocal folds close, the arytenoids tilt forward to contact the base of the epiglottis, the larynx elevates under the base of the tongue and the epiglottis tilts backward to seal the laryngeal vestibule. The opening of the upper oesophageal sphincter (UOS) is facilitated through the relaxation of the cricopharyngeous muscle, contraction of the suprahyoid and thyrohyoid muscles, and the pressure of the descending bolus.[6] The oesophageal phase is the final phase of the swallow, which begins as the bolus moves through the UOS, to be transported via automatic peristaltic waves to the stomach.[7]

Specific patterns of oral, pharyngeal and oesophageal impairments in feeding have been documented in children with CP. They may have difficulty in the oral phase of the swallow due to inadequate function of the oral muscles, exaggerated oral reflexes and altered oral sensitivity.[8] This may include limitations to tongue lateralisation necessary for chewing solids, excessive tongue thrusting, impaired bolus transit, increased oral transit time (greater than 3s) and reduced ability to clear food residue in the mouth. Poor control of the lips may result in difficulty receiving the bolus (eg, sipping from a cup or clearing a spoon), difficulty sucking from a bottle or straw, anterior loss of food due to poor lip seal and excessive saliva loss.[8] Children may also have pharyngeal phase impairments, including delayed or incomplete closure of the airway during the swallow, oropharyngeal aspiration of food or fluid and food residue in the pharynx.[9] Aspiration is defined as passage of material below the vocal folds.[6] This can be oropharyngeal aspiration (primary) of orally ingested material, saliva or mucous secretions; or reflux aspiration (secondary) of gastro-oesophageal refluxate. Aspiration can occur before the swallow (due to lingual disco-ordination allowing the bolus to prematurely spill over the base of the tongue, or a delayed swallow trigger); during the swallow (associated with ineffective laryngeal closure or disco-ordination); or after the swallow (related to laryngeal/pharyngeal residue falling into the reopened airway).[6] Usually food entering the laryngeal vestibule and subglottic space triggers a cough, which is a major protective mechanism of the airway.[6] Silent aspiration occurs when food or fluid enters below the true vocal folds with the absence of clinical signs or symptoms, which is commonly reported in children with CP.[9 10] Gastrointestinal impairments (including reduced motility and reflux) occur frequently in individuals with feeding problems and CP, both secondary to and contributing towards the difficulty.[11]

It is believed that OPD is highly prevalent in individuals with CP; however, there is a lack of comprehensive population-based data.[5 12–26] Estimates of prevalence vary significantly, from 19% in a large register sample,[24] to 99% in a sample of children with moderate–severe gross motor impairment.[14] Much of the literature exploring OPD in feeding has been limited by study methodology and case-definition of OPD. Many studies have based the prevalence of OPD on parent report or non-validated methods, and samples have generally been limited to individuals with more severe gross motor impairments[12 14 15 17] and across a broad age range.[5 12 14–22 26] The findings from key studies have been summarised in table 1.

Indirect or inconsistent means of OPD case identification have regularly been utilised in studies, with OPD identified through parent report,[12 13 15–17] chart reviews[5 17] and non-standardised assessments.[21–23 25] The variability in the method of case identification limits comparisons between these studies, and makes it difficult to estimate the true prevalence of OPD in the paediatric CP population. Parents have been shown to underestimate the presence of impaired feeding skills compared to formal clinical evaluation,[14] so prevalence data using these methods may represent an underestimate of the true population prevalence of OPD. Most parent questionnaires in the reported studies lacked adequate validity and reliability data, reducing confidence in these results.[12 13 15–17]

The generalisability of prevalence estimates of OPD to the general population of children with CP has been limited in most studies due to a focus on feeding skills in children with moderate–severe gross motor impairment.[12 14 15 17 25] Many of the studies which sampled across the range of gross motor severity have still had a disproportionate number of individuals from the more severe classifications.[5 16 18 20 21] This is largely due to

Table 1 Prevalence of oropharyngeal dysphagia in children with cerebral palsy and its relationship to gross motor function

Author and year	Participants	OPD measure	Gross motor measure	Major findings
Santoro *et al* (2012)	n=40 children with CP and feeding problems aged 4 months–11 years, GMFCS III–V	Parent questionnaire and mealtime observation by SP	GMFCS CP motor type	Children from GMFCS III showed best feeding performance (hemi/diplegic CP)
Erkin *et al* (2010)	n=120 children with CP, 2–18 years	Informal observations of feeding behaviours	GMFCS (collapsed to two groups) CP motor type	22% feeding dysfunction (12% mild, 8% moderate and 2% severe) Feeding dysfunction in 4% of GMFCS I–III, and 22% of GMFCS IV–V (p<0.001)
Parkes *et al* (2010)	n=1357 children with CP, median 5;11 years, GMFCS I–V	Question on standardised assessment for register ('absent' or 'present')	GMFCS CP motor type (Surveillance of CP in Europe Project)	19% chewing and swallowing problems GMFCS significantly related to swallowing/chewing difficulties and excessive drooling: GMFCS IV—OR 4.8 GMFCS V—OR 15.7
Wilson and Hustad (2009)	n=37 children with CP, 11–58 months (mean 41 months)	Parent report on feeding and swallowing Questionnaire Clinical evaluation of OPD (no formal tools)	No analysis of motor severity	56% had difficulty feeding from a bottle 78% had oral motor involvement (including motor speech) No analysis with gross motor
Ortega *et al* (2009)	n=53 children with CP, 3–13 years, GMFCS I–V (with 75% of sample from IV–V)	Oral Motor Assessment Scale	GMFCS	83% did not have functional feeding skills No analysis with gross motor
Calis *et al* (2008)	n=166 children with severe CP and ID, 2–19 years (mean 9;4 years). GMFCS IV–V, IQ<55	DDS and DSS Parent report	GMFCS	99% clinically apparent dysphagia Oral motor severity positively associated with motor functional severity (p<0.001) Postural stability positive association to DDS score, but not postural alignment for eating
Yilmaz, *et al* (2004)	n=23 children with spastic CP, 4–25 years GMFCS I–V	FFAm	Ambulatory status	50–74% normal–mild feeding difficulties; 30–51% moderate–severe feeding difficulties
Field *et al* (2003)	n=44 children with CP, 1 month–12 years (median age range 13–36 months)	Record review	No analysis of motor severity	68% oral motor delay 32% dysphagia
Fung *et al* (2002)	n=230 children with CP, 2–18 years (mean 9.7 years), GMFCS III–V	Parent reported on feeding questionnaire—rated as none, mild, mod and severe	GMFCS	48% feeding problems GMFCS level was highly associated with the degree of feeding dysfunction (p<0.001)
Sullivan *et al* (2000)	n=271 parents of children with childhood impairments (96% CP), 4–13 years, mild–severe gross motor	Register question to determine 'articulation/swallowing problems' Parent questionnaire to	Parent rated severity of motor function, relating to aids needed (mild, mod and severe)	79% articulation or swallowing problems Significant correlation between severity of gross motor impairment and

Continued

Table 1 Continued

Author and year	Participants	OPD measure	Gross motor measure	Major findings
		investigate specific feeding problems		range of specific feeding problems (eg, choking with food p<0.001; prolonged mealtime p<0.001)
Reilly *et al* (1996)	n=49 children with CP, 12–72 months, mild-profound (70% with severe-profound imp)	SOMA Early feeding histories	Standard Recording of Central Motor Deficit—classified as no disorder/mild; severe/profound	Positive relationship between OPD severity and gross motor severity (p=0.000) Mod and severe OPD more common in tetraplegia, whereas diplegia was associated with mild OPD (p=0.001)
Dahl *et al* (1996)	n=35 children with CP, 2.4–15.2 years (mean 7.7 years), profound motor handicaps (moderate and severe CP)	Parent interview (retrospective data of 4 weeks) triangulated with medical file review	Motor severity differentiated by level of dependence	60% reported as having daily feeding problems No analysis of gross motor
Stallings *et al* (1993)	n=142 children with quadriplegic CP, 2–18 years	Parent interview (0–5; 0=no problems, 5=all (5) oral motor problems)	Diagnostic criteria (for quadriplegic CP) not defined in paper	86% impaired oral motor ability No analysis of gross motor
Waterman *et al* (1992)	n=56 children with CP, 5–21 years (median 14 years), mild–severe	Chart review (clinical or radiographical dysphagia) Interviews with SP	Severity defined based on ambulatory status from chart review	27% had evidence of swallowing disorders More severe CP in dysphagic group ('consistent but non-significant trend'—no statistics reported)
Thommessen *et al* (1991)	n=42 children with CP, 1–16 years	OPD evaluated by 3 OTs/PTs (based on child's age)	No analysis of motor severity	33% had OPD No analysis of gross motor
Love *et al* (1980)	n=60 children with CP, 3–23 years (mean 12.5 years), spastic, athetoid and mixed; mild–non-ambulatory	Non-standardised oral-motor tasks (biting, sucking, swallowing, chewing soft and firm food)	No analysis of motor severity	40% with inadequate feeding

CP, cerebral palsy; DDS, Dysphagia Disorders Survey; DSS, Dysphagia Severity Scale; FFAm, Functional Feeding Assessment modified; GMFCS, Gross Motor Function Classification System; ID, intellectual disability; imp, impairment; mod, moderate; OPD, oropharyngeal dysphagia; OT, occupational therapist; PT, physiotherapist; SOMA, Schedule for Oral Motor Assessment; SP, speech pathologist.

sampling bias, with most studies recruiting from special schools or clinic databases, thus limiting the sample representativeness. In addition, a range of measures have been used to determine gross motor severity, including formal classification systems such as the Gross Motor Function Classification System (GMFCS), and criteria developed for the individual study. This limits our ability to accurately quantify the prevalence of OPD across the full range of gross motor severity, from mild to severe, and may provide an overestimate of the prevalence in the general population of children with CP if rates are extrapolated based on the moderate–severe sample.

Feeding skills develop rapidly in the early years as children transition through a range of food and fluid textures, related to their developing anatomy, neurology and physiology.[27] Rapid development of sensorimotor integration of swallowing and respiration, upper limb skills, posture and psychosocial maturation occur during the first 3 years.[7] By 18 months children are typically sitting independently, with fully co-ordinated swallow and respiration, and taking a full range of textures.[7] The development of chewing skills continues into childhood, with the adult co-ordination of lateral and vertical jaw movements emerging between three and 6 years.[28] Most prevalence studies of OPD in children with CP have been designed to examine oral sensorimotor skills in samples with a broad age range from early childhood (4 months to 4 years) through to adolescence or early adulthood (11–25 years).[5 12 14–19 21 26] The mean age for many of these studies was 9 years. Only two studies

limited their sample to preschool years, with participants ranging in age from 12 to 72 months[20] and 11 to 52 months.[13] Few children from the toddler or preschool age range have been sampled in previous studies, so a gap in knowledge remains. It is important to begin to delineate OPD in this critical age range to facilitate early identification and intervention, and to explore the progression of early feeding skills and their changing relationships with other associated factors (eg, growth, nutrition and respiratory health).

It is well accepted clinically that there is an interaction between an individual's oral sensorimotor skills in feeding and their gross motor skills. An individual's feeding posture can impact on their swallow by promoting poor alignment or reducing the stability for controlled oral movements, as well as the influence of the neurological lesion on all motor skills.[15 29] Poor head position has been related to compromised airway protection by opening the airway, and influencing the flow rate of foods/fluids swallowed.[30] The precise relationship between body position and swallow-breath coordination continues to be explored.[31] This relationship between OPD and gross motor skills is supported in the literature, with the prevalence and severity of OPD reported to be positively correlated with the extent of motor involvement.[5 14–16 20 24] However, these findings lack weight due to few studies using direct objective measures of oral sensorimotor skills,[5 14–16 23 24] a lack of validated measures of gross motor skills[5 16] or sampling only children with moderate–severe gross motor impairment.[14 15 25]

The Oxford Feeding Study of 271 children with OPD, found those with more extensive motor involvement, that is, quadriplegia and dyskinesia, were most likely to have difficulties with swallowing and articulation, based on parent report.[16] Those unable to walk or who required an aid and helper to walk were more likely to have problems eating and swallowing lumpy food, to need food mashed or liquidised, and were also more likely to be fed via a tube. In a large register-based study (n=1357), the odds of having swallowing/chewing difficulties and excessive drooling increased significantly as GMFCS level increased;[24] however, this study only used a single standardised question to determine the presence of feeding difficulty. Using validated assessments (Schedule for Oral Motor Assessment (SOMA) and Standard Recording of Central Motor Deficit categories), the presence of gross motor impairment was significantly associated with the presence of oral motor dysfunction in a cross-sectional community-based sample of 49 preschool children with CP.[20] While strengthened by using validated measures for both oral motor and gross motor skills, the sample was small and only used binary outcomes (presence/absence of dysfunction). The relationship between OPD and gross motor skill attainment will be strengthened by exploring this association across a number of gross motor severity levels using the GMFCS.

The feeding impairments resulting from OPD may impact negatively on many dimensions of an individual's health, including the child's development, growth and nutrition, chest status and respiratory health, gastrointestinal functioning and parent–child interactions.[32] Both OPD and tube feeding are demonstrated risk factors for increased premature mortality in individuals with CP.[33–35] Optimal nutrition in the early years forms a critical foundation for improved health across the lifespan. Compromised nutritional status influences children's mood and irritability, muscle spasticity, healing, peripheral circulation and general well-being.[36] In addition, OPD can result in acute and/or chronic oropharyngeal aspiration which is significantly associated with compromised respiratory status, including recurrent lower respiratory tract infections and chronic lung disease.[9 19] Understanding the nature and severity of OPD in young children with CP and its relationship to gross motor attainment, growth and nutritional status, will inform health interventions, benefiting children with CP and their families, and potentially lowering costs of healthcare.[37]

Aims and hypotheses

This study will investigate the relationship between OPD, gross motor skills, growth and nutritional status in young children with CP across two critical age points, 18–24 and 36 months, corrected age. Specifically, this study aims to:

1.
   A. Systematically review the literature determining the clinimetrics of measures of OPD in preschool children with CP.
   B. Test the psychometric properties of the SOMA, Dysphagia Disorders Schedule (DDS) and Pre-Speech Assessment Scale (PSAS) in young children with CP.
2.
   A. Determine the prevalence of OPD and its subtypes (impaired saliva control, oral phase impairment and pharyngeal phase impairment) in a population of children with CP at 18–36 months.
   B. Explore the nature of the relationship between OPD and gross motor functional severity (according to GMFCS levels); and growth and nutritional status.
3. Longitudinally examine the potential risk factors for OPD (including gross motor attainment, anthropometric measures, dietary intake, ingestion functions, food and fluid textures, gender, age and socioeconomic factors) in children aged 18–24 and 36 months with CP.

These aims will be explored through the following three hypotheses:

H_1: The SOMA and DDS will be the most valid and reliable measures of OPD in young children with CP. The PSAS will have the best clinical utility.

H_2:
- A. There will be a negative relationship between OPD prevalence and gross motor function in children with CP aged 18–36 months.
- B. There will be a positive relationship between OPD prevalence, poor growth and nutritional status in children with CP aged 18–36 months.

H_3: Gross motor function, poor growth and nutritional status will have a greater association with OPD in children with CP than demographic risk factors.

Study significance

The results of this study will:

- Determine the accuracy of the SOMA, DDS, PSAS and signs suggestive of pharyngeal phase impairment, in detecting and evaluating OPD in preschool-aged children with CP.
- Contribute population-based data on the prevalence of OPD and subtypes, in children with CP using standardised measures. To date there is limited comprehensive population data across all gross motor severity levels. These data are essential before intervention trials can be conducted.
- Delineate the relationship between OPD and gross motor skill attainment in children with CP. Greater understanding of this relationship will assist in proactive screening in early intervention services, including early detection of children at risk of aspiration and compromised chest status, and prevention of negative health effects.
- Further explore potential associations between OPD and nutritional status and growth in children with CP. This will allow for greater access to preventative nutritional treatments and the development of more targeted interventions, thus promoting growth and overall health outcomes in young children with CP.

METHODS AND ANALYSES

This prospective longitudinal cohort study aims to recruit 200 children with CP born in Queensland, Australia, between 1 September 2006 and 31 December 2009. The OPD study is part of a larger longitudinal population-based study, Queensland CP Child: Growth, Nutrition and Physical Activity, which is exploring growth, nutrition and physical activity in children with CP (National Health and Medical Research Council (NHMRC) Australia, 569605). This study is being conducted in conjunction with another study, Queensland CP Child: Motor Function and Brain Development Study (NHMRC 465128). Figure 1 visually represents the relationship between these studies and the OPD substudies, which include:

1. Validity and reproducibility studies
   - A. Discriminative validity with typically developing reference sample;
   - B. Convergent validity with an additional OPD measure;
   - C. Reproducibility (test-retest, intrarater, inter-rater).
2. Cross-sectional study of children aged 18–36 months
   - A. Overall prevalence of OPD, subtypes and association with gross motor;
   - B. Oral phase impairment;
   - C. Pharyngeal phase impairment;
   - D. Functional feeding skills on food and fluid textures.
3. Longitudinal study of children between 18–24 and 36 months.

Recruitment

State-wide subject recruitment started in April 2009 in collaboration with the Queensland Cerebral Palsy Register, the Queensland Cerebral Palsy League, the Royal Children's Hospital (RCH) Brisbane, the Queensland Cerebral Palsy Health Service, the Royal Women's Hospital Brisbane and the Mater Children's Hospital. Paediatricians, general practitioners, allied health professionals, child health nurses and neonatal follow-up clinics are encouraged to refer children with motor delay (not sitting at 10 months, not standing at 12 months or walking at 24 months) for confirmation of a diagnosis of CP at the RCH/Mater Mothers' Hospital Specialist clinics. High ascertainment is expected for children across all levels of motor severity (GMFCS I–V) particularly as many of these children access services through the Queensland Cerebral Palsy Health Service, one of the key referral sources. Children who are detected after 18 months of age will be entered into the study later, at the time of diagnosis. Children can enter the study at 18, 24, 30 or 36 months age points. Those entering at 18 or 24 months will have their second assessment point collected at 36 months, and will be included as part of the longitudinal study. Children entering at 30 or 36 months will have their second assessment at 48 months, and therefore will not be included in the longitudinal study detailed in this study protocol. Further details of study entry and feasibility can be found in the larger study's protocol.[38]

Forty children with typical development aged 18–36 months (stratified for age) will be recruited to participate as a reference sample for the study. Siblings of children participating in the overall study will be invited to participate, as well as recruitment through staff newsletters, a hospital childcare centre and participants from other studies within the centre.

Selection criteria

Inclusion criteria

Children aged 18–36 months corrected age at the time of evaluation (birth-years 2006–2009), born in Queensland, with a confirmed diagnosis of CP are invited to participate in the present study. For the present study, CP is defined as a disorder of movement and/or posture and motor function, which must be permanent but not unchanging, and due to a non-progressive

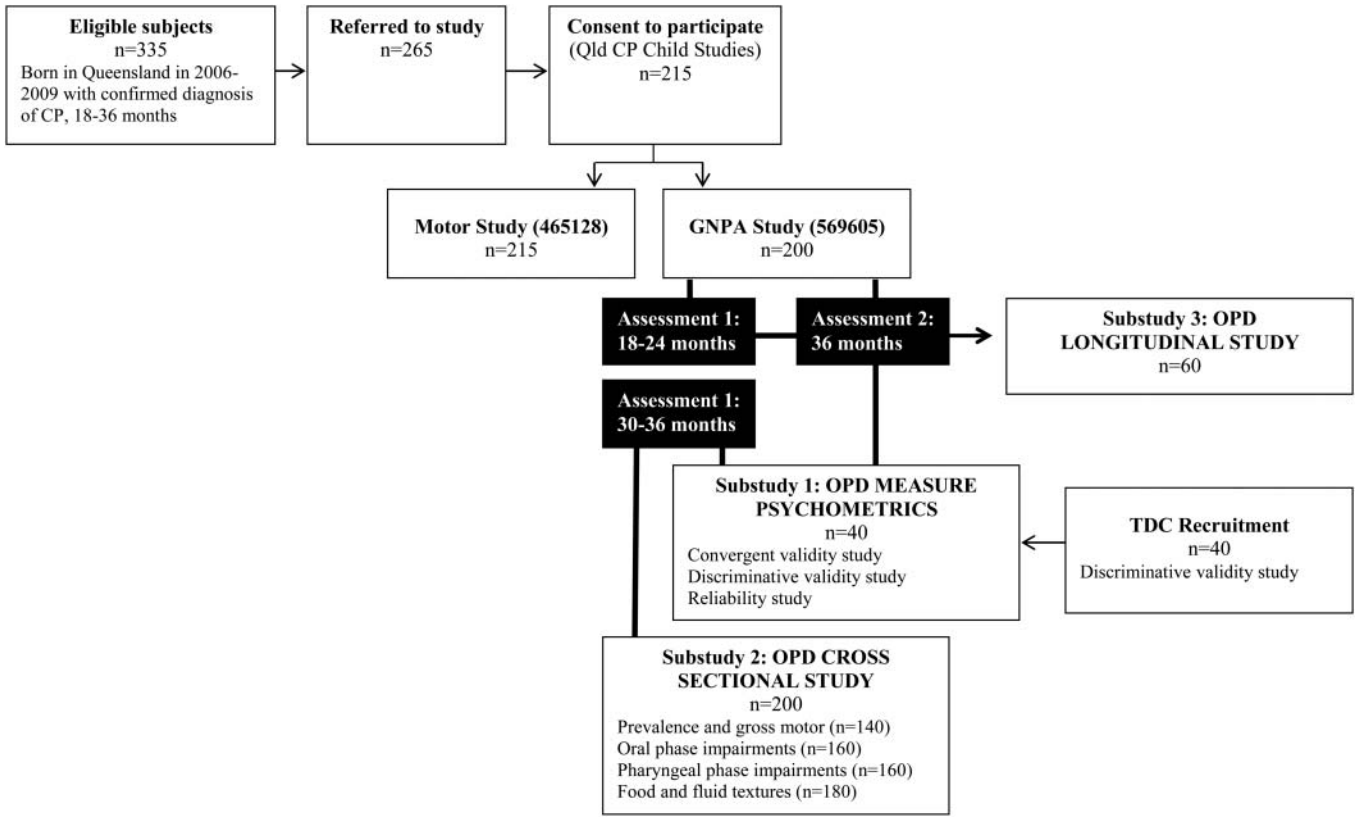

Figure 1 Critical pathways for oropharygeal dysphagia study. CP, cerebral palsy; GPNA, growth nutrition and physical activity; OPD, oropharyngeal dysphagia; Qld, Queensland; TDC, typically developing children.

interference/lesion in the developing brain (congenital lesions only).[4] The characteristic motor types are spasticity and dyskinesias (ataxia, rigidity and dystonia), and clinical features may also include negative signs of the motor neurone syndrome (muscle weakness and poor selective motor control).[39]

Exclusion criteria

Children diagnosed with a progressive or neurodegenerative lesion and children born outside Queensland are excluded from the study.

Typically developing reference sample

Children are eligible to participate in the reference sample if they are aged 18–36 months; born full term (<37 weeks); with no admissions to neonatal care, no diagnosis receiving medical or allied healthcare; and not on regular medications.

Measurements and procedures

Following confirmation of a diagnosis of CP, children attend the RCH for an assessment session with their family. During this visit, children are assessed using the Gross Motor Function Measure (GMFM),[40] Manual Ability Classification System (MACs),[41] anthropometric measurements taken, questionnaires administered to the parent/caregiver verbally (including the Pediatric Evaluation of Disability Inventory,[42] and Queensland CP Child: Growth, Nutrition and Physical Activity: Feeding Questionnaire) and the child's mealtime is videotaped.

Children participating in the reproducibility study will be invited to return to the hospital within a month to have a repeat mealtime video. If this is not possible, a home visit will be conducted. Children participating in the typically developing reference sample will be assessed at the hospital or at home, for a single mealtime video.

Feeding evaluation

During the feeding assessment, the child is well positioned in their typical mealtime seating (ie, chair, stroller and carer's arms). The video camera is set up to include a view of the child's face and neck, angled to the side of the feeder's shoulder of the hand that is not feeding the child, as per the study snack protocol. Prior to and following the mealtime, the researcher videoing the session records observations regarding clinical swallow signs (wet/gurgly voice, wet/gurgly breathing, rattly chest or the presence of cough) and severity of drooling. These ratings are confirmed by the speech pathologist when rating the videos. During the video session, the child is given three standardised presentations of each of four textures (puree, lumpy, chewable and fluid) by their primary carer, as outlined in the SOMA administration manual.[43] Purees include foods such as yoghurt, mousse or pureed fruit. Lumpy foods

could include semisolid (eg, baked beans, roughly mashed vegetables) or solid foods (eg, fruit salad) from a spoon. For the purpose of this assessment, chewable foods are items that are finger fed, usually requiring biting, including biscuits or whole fruit. Following these standard presentations, the child is allowed to complete the snack eating independently or assisted by their primary carer.

Primary measures

A major limitation in studies of OPD is the lack of widely accepted, validated and reliable measures.[15] The aim of the present study is to gather information regarding OPD that reflects children's performance in naturalistic environments (eg, home and childcare centres). For this reason, non-invasive observational methods were selected as part of the standard protocol for all children. The SOMA,[44] Dysphagia Disorders Survey: Pediatric (DDS),[45] and PSAS[46] were selected through systematic review as the most appropriate non-invasive objective clinical measures for the detection of dysphagia for this study.[47] The video tapes of children's mealtimes are formally rated by an independent speech pathologist, and data recorded using the standard assessment forms. Sixteen clinical pharyngeal signs suggestive of aspiration are also rated for each food/fluid texture, in conjunction with the rating completed in the session. The use of videos in mealtime observations is recommended in the SOMA administration manual to allow repeated viewing for more accurate description of motor tasks. The speech pathologist is certified in the use of the DDS to meet the validation standards.[48] The allocation of GMFCS level is masked to the speech pathologist when rating mealtime videos. If clinically indicated, some children have further evaluation of their OPD using instrumental assessments, such as Video Fluoroscopic Swallow Study (VFSS). This information is collected when available but is not part of the standard protocol for all children.

Schedule for Oral Motor Assessment

The SOMA is a standardised discriminative assessment which quantifies OPD in children aged between 8 and 24 months.[43] It was originally designed to evaluate children with no/mild neurological dysfunction, but subsequently was used to evaluate oral motor dysfunction (OMD) associated with a number of causes including neurological impairments.[43] The tool categorises children as OMD or normal oral motor function based on specified thresholds for each of seven oral motor challenge categories (OMCC) (puree, semi-solid, solid, cracker, bottle, trainer cup and cup).[43] The tool is predominantly a test of oral phase dysfunction; however, some items pertain to swallowing and the pharyngeal phase. Children are only scored on food/ fluid textures they accept during the assessment. The standardised administration of textures outlined in the administration manual is maintained in this study as much as possible, while allowing some flexibility for individual child and family factors to optimise the naturalistic context of the assessment.

The SOMA has been validated on 127 young infants; 58 comparison children with typical oral skills, 56 with non-organic failure to thrive (aged 8–24 months), and 13 children with CP and overt feeding difficulties (aged up to 42 months).[49] The abnormality score (total number of OMCCs with OMD) for children with CP was significantly different from the comparison group ($p<0.0001$). Individual OMCCs do not have adequate discriminative validity reported to be analysed as individual subtests, with 8–77% false negatives in the CP group.[49] The reliability of the measure was established by two independent speech pathologists rating three trials of 10 randomly selected videos from the sample. It has strong inter-rater reliability ($\kappa=1.0$ in 68% of fluid category items and 58% of food category items) and test–retest reliability between boluses ($\kappa=1.0$ in 84% of items).[50]

Dysphagia Disorders Survey

The DDS was developed as an evaluative screening tool to assess feeding and swallowing function in children and adults with a developmental disability.[48] Through observation of a typical mealtime, it identifies those with signs of oral preparation, oral initiation, pharyngeal and oesophageal phase dysphagia.[48] The measure is divided into two distinct parts: Part 1 scores dysphagia-related consequences (such as low weight, adaptive utensils and position); Part 2 rates the specific oral functions observed across three textures (non-chewable food, chewable food and fluid). The raw score from Part 1, and percentiles which are derived from both Parts 1 and 2, are not used in this study as they assess consequences of mealtime difficulty rather than specifically OPD. Part 2 provides a raw score that indicates an individual's functional eating competency (with a maximum impairment raw score of 22) and has been used previously as a measure of OPD.[14]

The DDS underwent final standardisation on 427 individuals with mean age of 33 years.[14] The paediatric measure was developed in a group of 166 children (range 2 years 1 month–19 years 1 month; mean 9 years 4 months), with moderate-severe CP (GMFCS III-V) and intellectual disability.[14] Test validity and interitem reliability were derived from an initial sample of 626 people with developmental disability.[51] Convergent validity was demonstrated in two studies comparing DDS scores to blinded speech pathologist diagnosis.[14 48] Inter-rater reliability of 97% agreement was calculated from a sample of 21 participants rated by six speech pathologists (each pair of speech pathologists rated seven participants).[48]

Dysphagia Severity Scale

The Dysphagia Severity Scale was developed by Calis *et al*[14] to provide a severity rating from the DDS Part 2 raw scores. Individuals are classified as one of the four severity levels, with level one being no disorder, and level four a profound disorder. The mild classification

and moderate–severe classification are differentiated by the presence of pharyngeal phase impairments (items 13–14 on the DDS), in addition to a score of one or more on the DDS Part 2. A profound disorder is reflected by non-oral status of individuals due to the severity of their OPD.

Pre-Speech Assessment Scale

The PSAS is an evaluative measure that examines 27 pre-speech feeding behaviour performance areas related to sucking, swallowing, biting, chewing, respiration-phonation and sound play.[46] It is appropriate for use with children with a neurological impairment, as well as those with typical development. Each subtest is scored on an ordinal abnormality scale (1–9) and a developmental scale (with age norms to 24+ months), to provide a double score overall. This provides comprehensive information on both dysfunctional and delayed feeding behaviour expected up to 24+ months.

The PSAS was developed through a 3-year longitudinal study of six children, and field testing of the measure for 8 years by 215 trained clinicians who provided annual feedback on its clinical use.[46] Other aspects of the measure's validity have not been tested. Reliability has been shown to be strong, although only in two studies with limited methodology.[46 44] Intra-rater reliability was 96% for 25 feeding behaviours which were scored in the six typically developing children.[44] Inter-rater reliability for this same sample was similarly excellent between two raters (92%).[44] Inter-rater reliability was fair to good when rated from video footage, with 65–87% agreement when 75 clinicians' ratings were compared to a predetermined standard of correctness for 78 children.[46]

Signs suggestive of pharyngeal phase impairment

Premealtime and post-mealtime observations of the presence or absence of (1) wet/gurgly voice (2) wet/gurgly breathing, (3) rattly chest and (4) cough are rated face-to-face in the mealtime session by a trained research assistant, to assess clinical signs of pharyngeal phase difficulty. A determination of pharyngeal phase impairment is noted if a child demonstrates any one of these signs, or 1 of 16 signs rated from video by the speech pathologist. These behaviours include gagging, coughing, choking, vomiting, throat clearing, multiple swallows, wheezing, stridor, rapid or laboured breathing, gurgly voice, rattly chest, snuffly nose, eye tearing, circumoral cyanosis/duskiness and food refusal and are noted for each food and fluid texture. These signs were selected from the literature[10 45] and research conducted by one of the investigators (KAW).[52]

A cross-sectional study of 150 children with dysphagia (mean age 16 months) compared retrospective data of pharyngeal phase impairments identified by VFSS to 11 commonly reported clinical signs and symptoms to determine their sensitivity and specificity.[52] Wet voice (sensitivity 0.67 and specificity 0.92), wet breathing (sensitivity 0.33 and specificity 0.83) and cough (sensitivity 0.67 and specificity 0.53) were considered good clinical markers of oropharyngeal aspiration on thin fluids, but not for puree textures.

Thomas-Stonell & Greenburg Scale—saliva control

The Thomas-Stonell & Greenberg[53] Scale is a semiquantitative assessment of drooling severity (one-point to five-point scale of no drooling to profuse drooling) and frequency (one-point to four-point scale of no drooling to constant drooling). A pre- and post-mealtime severity rating is recorded by trained researchers within the mealtime assessment and confirmed by the speech pathologist from video. In addition, a severity and frequency rating by the parents is collected based on observations during the previous week, and information reporting on the representativeness of this rating.

In a case–control study of 14 children with saliva loss and spastic CP aged 7–18 years (mean 11;7 years), drooling frequency and severity were reported by parents on the Thomas-Stonell & Greenberg Scale.[54] A Drooling Quotient, derived from parent scores, was compared to a more objective measure of weighing saliva loss on bibs and shown to be positively correlated (Spearman's $r=0.604$ $p<0.05$).[54]

Gross Motor Function Classification System

The GMFCS is a five-level classification system of children's functional gross motor severity. It is based on self-initiated movements, anti-gravity postures and motor skills expected in a typical 5-year-old children.[55] Children who are independently ambulant are classified as GMFCS I or II, those requiring an assistive mobility device to walk classified as GMFCS III and those in wheeled mobility as GMFCS IV and V. Two physiotherapists, trained in the use of the GMFCS, independently observe and classify children in one of five functional categories.[55]

The GMFCS has internationally established validity, reliability and stability for the classification and prediction of motor function of children with CP aged 2–12 years.[55–57] It has a high inter-rater reliability (generalisability=0.93).[56] Classification of gross motor abilities change with age, therefore separate descriptions are used for different age bands. In the current study, the <2 and 2–4 years descriptions are used. Lower inter-rater reliability is documented for the <2 years age band ($\kappa=0.55$), as younger children's gross motor abilities are more variable, and less developmental information is available on which to base the classification .[58] Test–retest reliability from <2 to 12 years appeared to be acceptable (generalisability coefficient=0.68). The GMFCS has been correlated with a number of motor scales, as well as CP distribution and type of motor impairment.[59]

Anthropometry

Height or length (depending on children's ability to stand) is measured to the last completed millimetre by a portable stadiometer/length board (Shorr Productions,

Maryland, USA). Where a direct measure of height or length is not possible, height is estimated using published equations from knee length or upper-arm length[60] measured with an anthropometer (Holtain Ltd, Dyfed, UK). Weight (measured to the nearest 100 g using chair scales; Seca, Germany) and skin-fold thickness (tricep and subscapular skinfolds, measured in millimetres with Harpenden callipers (Holtain Ltd)) measures are taken and body mass index (BMI) calculated (as weight/height, m^2) to assess children's nutritional status. Skin-fold measurements and BMI will be converted to z scores for analysis.[61] All measures are conducted by trained investigators. Full details of anthropometric procedures are provided in the larger study protocol paper.[38]

Dietary intake

A 3-day-weighed food record is used to measure children's typical dietary intake.[62] Parents are instructed on the standard protocol to ensure accuracy and consistency in completing the food record. Food records will be analysed for the percentage of children's diet made up of food and fluid textures. Food records are also analysed using the Foodworks dietary analysis software program (Xyris Software (Australia) Pty Ltd, Kenmore Hills, Australia) to give information regarding energy, carbohydrate, fat and protein intake.

Secondary measures

Queensland cerebral palsy child: growth, nutrition and physical activity: Feeding Questionnaire (Qld CP Child Feeding Questionnaire)

The Qld CP Child Feeding Questionnaire gathers parent report on their child's oral sensorimotor and mealtime function. Parent report will be used to triangulate findings from clinical assessment to gain a more comprehensive picture of the child's skills across settings and time. It includes:

- Severity and frequency of saliva loss using the Thomas-Stonell & Greenberg Scale (above).
- The impact of saliva on four domains, including the impact on child and family measured using a 10-point visual analogue scale.
- Types of food and fluid included in the child's diet: inclusion of textures rated for four fluid levels (thin, mildly thick, moderately thick and extremely thick) and five food textures (puree, thick puree, lumpy mashed food, chewable solids and tough chewable foods). Fluid terms align with the Australian Standardised Labels and Definitions.[63]
- Presence of eating or drinking problems: rated on a four-point scale from no feeding problems to severe difficulties. Severity is also rated for eating and drinking on a 10-point visual analogue scale.
- Mealtime behaviours and signs suggestive of pharyngeal phase impairment or aspiration are documented by parents against the same 16 signs and symptoms suggestive of pharyngeal phase impairment as is noted in clinical observation. Presence or absence of specific signs and symptoms were noted on each texture (thin fluid, thick fluid, puree, lumpy and finger foods).

Gross Motor Function Measure

Gross motor function is evaluated at each assessment using the GMFM (GMFM-66 and GMFM-88).[40] The GMFM is an evaluative tool that covers five gross motor domains, including lying and rolling; sitting; crawling and kneeling; standing; and walking, running and jumping. The GMFM-66 is a subset of items from the GMFM-88, developed through Rasch analysis, and is shown to be valid and reliable in children with CP.[64] The GMFM-66 will be used to provide an overall measure of gross motor function, and the GMFM-88 domain scores to explore specific motor skills. Scores are expressed as a percentage of the maximum score, which are skills expected of a typically developing child at 5 years.[65] The GMFM is not valid for comparisons of children across different age ranges, therefore all analyses using GMFM scores are completed in 18–24 and 30–36 months age brackets. Gross motor assessment is completed by two experienced paediatric physiotherapists who have criterion rating with the study developers (RNB).

Motor type and distribution

The type of CP (spastic, dyskinetic and hypotonic) and motor distribution (hemiplegia, diplegia and quadriplegia) is classified according to the Surveillance of CP in Europe.[66] This is assessed by two independent physiotherapists at each assessment.

Manual Ability Classification System

Children's manual ability is classified during performance in everyday activities according to the MACs. The MACs classifies children on a five-level scale based on how they use their hands when performing activities such as eating, dressing, playing and drawing.[41] This classification was developed for children aged 4–18 years, but has been shown to have good reliability for use in children as young as 2 years.[67] Children are rated by two independent physiotherapists.

Sample size calculations

Queensland cerebral palsy child: growth, nutrition and physical activity

On the basis of a reported incidence of CP of 2/1000 live births within Australia, there is an estimated 100 new cases of CP in Queensland each year.[3] For sample size calculations, a population prevalence estimate of 90% was taken from the study by Reilly *et al*.[20] In order to estimate the true prevalence of OPD in the population of children with CP with 95% confidence, a minimum sample of 35 participants were needed to provide sufficient precision within ±10% of the true value.

Owing to the limited data reported in the literature of prevalence based on direct clinical evaluation in the mild gross motor level, children in GMFCS I were hypothesised to have normal feeding skills. Nearly all

Table 2 Summary of primary outcome and exposure variables in the present study by objective and statistical tests

Hypothesis	Outcome variable	Exposure variable	Statistics
H2(A)	OPD overall (yes on SOMA, DDS, PSAS or clinical pharyngeal signs) *Dichotomous*	GMFCS GMFM-88 domains MACs Motor type/distribution	Prevalence, χ^2 Binomial logistic regression
H2(a)	SOMA (overall) *Dichotomous* DDS (overall) *Dichotomous* PSAS (overall) *Dichotomous* Pharyngeal signs (overall) *Dichotomous* Saliva control (overall) *Dichotomous*	GMFCS GMFM-88 domains MACs Motor type/distribution	Prevalence, χ^2 Binomial logistic regression
H2(A)	DDS Part 2 raw score *Continuous* Dysphagia Severity Score *Ordinal*	GMFM-66 GMFCS	Linear regression Multinomial logistic regression
H2(B)	Growth (height/length, knee and upper arm length) Ax1 *Continuous*	OPD and subtypes Ax1	Linear regression
H2(B)	Nutritional Status (skin-folds, BMI) Ax1 *Continuous*	OPD and subtypes Ax1	Linear regression
H3	OPD, SOMA, DDS, Pharyngeal Signs, Saliva Control, Parent Report Ax2 *Dichotomous*	OPD, SOMA, DDS, PSAS, Pharyngeal Signs, Saliva Control, Parent Report Ax1 *Dichotomous*	χ^2 to compare prevalence Binomial logistic regression
H3	OPD at Ax1 *Dichotomous*	GMFCS (collapsed)	Binomial logistic regression
H3	OPD at Ax2 *Dichotomous*	GMFCS (collapsed)	Binomial logistic regression
H3	Nutritional Interventions (tube feeding and/ or supplements) Ax2 *Ordinal*	OPD and subtypes Ax1	Multinomial logistic regression
H3	Growth (height/length, knee and upper arm length) Ax2 *Continuous*	OPD and subtypes Ax1	Linear regression
H3	Nutritional Status (skin-folds, BMI) Ax2 *Continuous*	OPD and subtypes Ax1	Linear regression

Ax1, 18–24 months assessment; Ax2, 30–36 months assessment; BMI, body mass index; DDS, Dysphagia Disorders Survey; GMFCS, Gross Motor Function Classification System; GMFM, Gross Motor Function Measure; MACs, Manual Ability Classification System; OPD, oropharyngeal dysphagia; PSAS, Pre-Speech Assessment Scale; SOMA, Schedule for Oral Motor Assessment.

children in GMFCS V have been reported to have OPD.[14] With an expected 40 participants per GMFCS level (total n=200), this study will be able to detect a significant difference between groups (80% power, α=0.05) if the true proportion of OPD in the population differs by >25% between groups.

Validity and reproducibility studies

Oropharyngeal dysphagia reproducibility study

With an expected agreement of greater than 90%, a sample of 20 children with CP per age band (a total of 40 children across 18–36 months age range and gross motor severity levels) will be able to give sufficient statistical power, with 95% confidence.

Oropharyngeal dysphagia discriminative validity study

In order to estimate the true mean score of typically developing children aged 18–24 and 30–36 months on the SOMA and DDS with 95% confidence (and precision of 0.5 around the estimate), a reference sample of 16 typically developing children from each age band (ie, n=16 18–24 months corrected age; n=16 30–36 months corrected age) will be needed. In total, we propose to recruit 40 children aged 18–36 months.

An estimate of the standard deviation of 0.3 for the typically developing group was based on a previous sample of typically developing children participating in the GNPA study aged 4 years (scored on the DDS). It is expected that the variability in the younger age range will be greater than the 4-year-old sample, and therefore a standard deviation of 0.5 was used to ensure that the sample is large enough to give precision to the estimate of mean scores. The DDS is the measure expected to have the greatest variability in scores, and therefore it has been used for the sample size calculations.

Statistical considerations

This study explores the relationship between OPD as an outcome variable (overall, impairment in saliva control, oral and pharyngeal phases and food/fluid textures) with the primary exposure variable of gross motor skill attainment. It also investigates OPD as an exposure variable for the outcomes of growth and nutritional status. The statistical analysis plan is summarised in table 2. Demographic data of the sample will be presented with descriptive statistics, and sample representativeness to the population determined by comparing the prevalence of GMFCS classifications to the non-participants and data reported in an Australian register study.[68]

Inter-rater and intrarater reliability of the primary measures (SOMA, DDS, PSAS, pharyngeal signs, saliva control and GMFCS) will be assessed using Cohen's Kappas

(weighted and unweighted), and percentage agreement will be used. Existing cut scores for the SOMA, DDS and PSAS will be evaluated for their sensitivity and specificity to accurately identify typically developing children as having no oropharyngeal dysphagia. The mean score of the typically developing reference sample ($Mean_{TDC}$)+two SD will be used to determine more appropriate cut scores for the measures (ie, scores above two SD of the $Mean_{TDC}$ are considered to indicate the presence of oropharyngeal dysphagia). The reference sample will be included in regression analyses for the overall study as a base group for comparison.

The strength of relationship between outcome and exposure variables will be analysed using regression modelling with ORs (for binary outcome variables) and relative risk ratios (for ordinal outcome variables). The 95% CIs will be calculated for all effect estimates. GMFCS levels will be collapsed into three groups (GMFCS I–II, GMFCS III and GMFCS IV–V) for regression models in the longitudinal study (n=60) to increase statistical power. All demographic data, such as age, gender and geographical location, will be used in regression models to explore potential confounding with the primary variables. Postcode will be used to allocate children into five geographical categories from highly accessible to very remote.[69] Likelihood ratios will be used to evaluate the influence of covariates on the models, using backward stepwise elimination. If a group within a model has perfect prediction of the outcome, ORs will be calculated after applying a continuity correction of 0.5 to each appropriate cell. All data analyses will be performed using Stata Statistical Software.[70] For all tests, significance will be set at $p<0.05$.

ETHICS AND DISSEMINATION

Ethics committee approvals have been gained through the University of Queensland Medical Research Ethics Committee (2008002260), the Children's Health Services District Ethics Committee (HREC/08/QRCH/112), the Mater Health Services Human Research Ethics Committee (1520EC), the Cerebral Palsy League of Queensland (CPLQ 2008/ 2010 1029), Gold Coast Health Service District Human Research Ethics Committee (HREC/09/QGC/88), Central Queensland Health Services District Human Research Ethics Committee (SSA/10/QCQ/13) and the Townsville Health Service District Human Research Ethics Committee (HREC/09/QTHS/96). There are no known health or safety risks associated with participation in any aspect of the described study. All families will give written informed consent to participate, and they are able to withdraw their child from the study at any time without explanation, without any penalty from staff at the Royal Children's Hospital or University of Queensland, or any effect on their child's care. Data collected in this study will be stored in a coded reidentifiable form (by ID number). Each child has three assessment appointments across the duration of the larger study, which necessitates data to be reidentifiable.

To our knowledge, this protocol outlines the first large population-based study using direct clinical feeding assessment in young children with CP. The results of this study are planned to be published in peer reviewed medical and clinical journals, and presented at relevant international conferences. The following publications are proposed:

- Validity and reproducibility of measures of orophayrngeal dysphagia for young children with CP.
- Oropharyngeal dysphagia in young children with CP and its relationship to gross motor skills.
- Oral phase impairment in young children with CP.
- Pharyngeal phase impairment in young children with CP.
- Functional feeding skills, food and fluid texture inclusion in diets of young children with CP.
- Longitudinal relationships between orophayrngeal dysphagia, gross motor skills, growth and nutritional status in young children with CP.

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Contributions KAB, KAW, KLB and RNB contributed to the study protocol. PSWD, RNB, KLB, RSW and KAW contributed to study concept, design and grant writing. All authors read, critically revised and approved the final manuscript.

Funding This work was supported by the National Health and Medical Research Council (NHMRC) Postgraduate Medical and Dental Scholarship (1018264—KAB), NHMRC Career Development Fellowship (APP1037220—RNB) and NHMRC Research Grants (569605 and 465128).

Competing interests None.

Ethics approval University of Queensland Medical Research Ethics Committee (2008002260).

Provenance and peer review Not commissioned; externally peer reviewed.

Data sharing statement Further details of the study protocol can be requested from the corresponding author.

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Longitudinal cohort protocol study of oropharyngeal dysphagia: relationships to gross motor attainment, growth and nutritional status in preschool children with cerebral palsy

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BMJ Open 2012 2:
doi: 10.1136/bmjopen-2012-001460

Correction

Benfer KA, Weir KA, Bell KL, *et al*. Longitudinal cohort protocol study of oropharyngeal dysphagia: relationships to gross motor attainment, growth and nutritional status in preschool children with cerebral palsy. *BMJ Open* 2012;**2**:e001460. A number of author corrections were inadvertently missed during the proofing stage:

1. The title of this paper should read: 'Protocol for a longitudinal cohort study of oropharyngeal dysphagia: relationships to gross motor attainment, growth and nutritional status in preschool children with cerebral palsy.'
2. Under the section "Aims and hypotheses" the expansion of DDS is actually "Dysphagia Disorders Survey" (not "Schedule").
3. Under the section "Thomas-Stonell & Greenburg Scale—saliva control", paragraph 2, reference 53 should be after "Scale" (as this is part of the measure name).
4. Table 2: H2(A) –These are all the same hypothesis, so all instances should have been in upper case A.

We apologise for these errors.

BMJ Open 2012;**2**:e001460corr1. doi:10.1136/bmjopen-2012-001460corr1

Update to Literature Review

Since the publication of the protocol paper in 2012, there were 7 new studies published on the prevalence and patterns of OPD/ feeding in children with CP, as shown in Table 4. Studies are increasingly using GMFCS as a means to describe the motor severity of their samples, which is helpful for beginning to compare the data. The cross-sectional study by Kim and colleagues clearly presented their OPD findings according to GMFCS level. They assessed 14 ingestion functions using VFSS, however their sample was only small (n=29).[72] Studies continue to use informal measures of OPD,[86-88] and 2 studies using VFSS (an objective measure of aspiration) did not use standardised protocols to evaluate other ingestion functions.[72,89] One study recruited only children with feeding impairments (n=118), therefore overall prevalence cannot be extrapolated.[89]

A special supplement of the *European Journal of Clinical Nutrition* was published in 2013 (volume 67, S1-S2), titled “A Practical Approach to the Nutritional Management of Children with Cerebral Palsy”. This supplement sought to update the scarce and out-dated guidelines surrounding this topic by gathering international experts at a meeting in 2012. An article in this supplement by Arvedson reviewed the types of OPD in children with CP, the importance of multidisciplinary assessment of feeding problems, and points to consider in management.[5]

Table 4. Findings Related to Prevalence of Oropharyngeal Dysphagia in Children with Cerebral Palsy and its Relationship with Gross Motor Function

Author & Year	Participants & Sampling	OPD Measure	GM	Major Findings
Lopes et al (2013)[88]	n=90 children with CP aged 2-12.8 years. Convenience sampling through rehab centre	Informal: parent report (chewing & swallowing)	Motor type and distribution	26.0% chewing impairment and 9.0% swallowing impairment overall; No impairment in children with diplegia; 41.0% chewing impairment and 12.8% swallowing impairment in children with tetraplegia; 14.5% chewing impairment and 6.6% swallowing impairment in children with hemiplegia.
Kim et al (2013)[72]	n=29 children with CP aged 2.5-16 years, GMFCS I-V. Recruited through hospital clinic	VFSS Parent questionnaire	GMFCS	I-II: 30.0% oral-preparatory impairment, 70.0% oral phase impairment, 60.0% pharyngeal phase impairment III: 71.4% oral-preparatory impairment, 100.0% oral phase impairment, 100.0% pharyngeal phase impairment IV-V: 100.0% oral-preparatory impairment, 91.7% oral phase impairment, 91.7% pharyngeal phase impairment
van den Engel-Hoek et al (2013)[89]	n=118 (53 with spastic CP, 34 with dyskinetic CP, 31 with neuromuscular disorders) aged 11 mth-19 years. Recruited through hospital clinic, VFSS eligible	VFSS (thin fluid & puree)	Motor type (spastic and dyskinetic)	All children with CP had OPD. Children with spastic CP had more piecemeal deglutition than those with dyskinetic Children with CP had significantly more anterior loss, pooling in valleculae, nasal regurgitation, laryngeal penetration and aspiration than neuromuscular

Author & Year	Participants & Sampling	OPD Measure	GM	Major Findings
				group.
Weir et al (2013)[90] [a]	n=170 children with CP aged 18-36 mths, GMFCS I-V. Population-based	Capability on food textures on PEDI	GMFCS	Capability to eat *cut-up chunky* and *all textures* of table foods decreased with increasing GMFCS ($P < .05$)
Martinez-Biarge et al (2012)[86]	n=126 children with basal ganglia injuries (n=89 with CP) aged 12-48 mths. Recruited from hospital MRI database	Informal observations (poor suck, spoon for liquids, cough/ splutter/ choke, long feeds, difficulty swallowing lumpy	GMFCS	65.0% feeding difficulties (half mild-moderate, half required tube feeds) Feeding impairment significantly related to GMFCS level ($P < .001$)
Dahlseng et al (2012)[87]	n=661 children with CP. Recruited from CP Register	5-point scale: level of independence in self-feeding/ tube feeding	GMFCS	21.0% completely dependent feeders/ tube-fed: significantly related to motor type ($P < .001$), GMFCS ($P < .001$) and fine motor function ($P < .001$)
Santos et al (2012)[91]	n=43 children with spastic CP aged 11-19 years. Recruited through rehabilitation institute	OMAS (categories collapsed into 2 groups: passive/ subfunctional and functional/ semi-)	No analysis	48.8% children were classified subfunctional and 51.2% classified as functional

Abbreviations: CP, Cerebral Palsy; GM, Gross Motor assessment; GMFCS, Gross Motor Function Classification System; OMAS, Oral Motor Assessment Scale; PEDI, Pediatric Evaluation of Disability Inventory; VFSS, Videofluoroscopic Swallow Study

[a] Participants in the study by Weir et al were part of the Growth, Nutrition and Physical Activity Study (the overarching study of this doctoral research)

Change to Proposed Recruitment Numbers

The target recruitment for the main GNPA cross-sectional study (aged 18 to 36 months) was reported as n=200 in the protocol paper (based on 60% recruitment across 3 full birth years and a partial recruitment year).[92] Children were able to enter the study at any 6-monthly interval between 18 to 60 months. Recruitment to the overall study at any age has been consistent with proposed 60% recruitment rate; however a notable proportion of children have entered after their third birthday (outside of the doctoral research eligibility age), resulting in lower recruitment than originally proposed. The final recruitment numbers are shown in Figure 4. This lower recruitment has not had significant implications for the study. The overall sample size of this study was still substantial (n=130) and adequate to provide precision to our overall prevalence estimates. Furthermore, much of the comparison between GMFCS levels has been with reference to the sample of children with TD (n=40), which was not in the original sample size calculation. Based on an estimated 25% of children from the TD sample being identified as having OPD, and prevalence estimates derived from the initial article,[93] the study was calculated to have sufficient power >0.97 (α=0.05). An overview of the number of participants in each published paper is shown in Table 5.

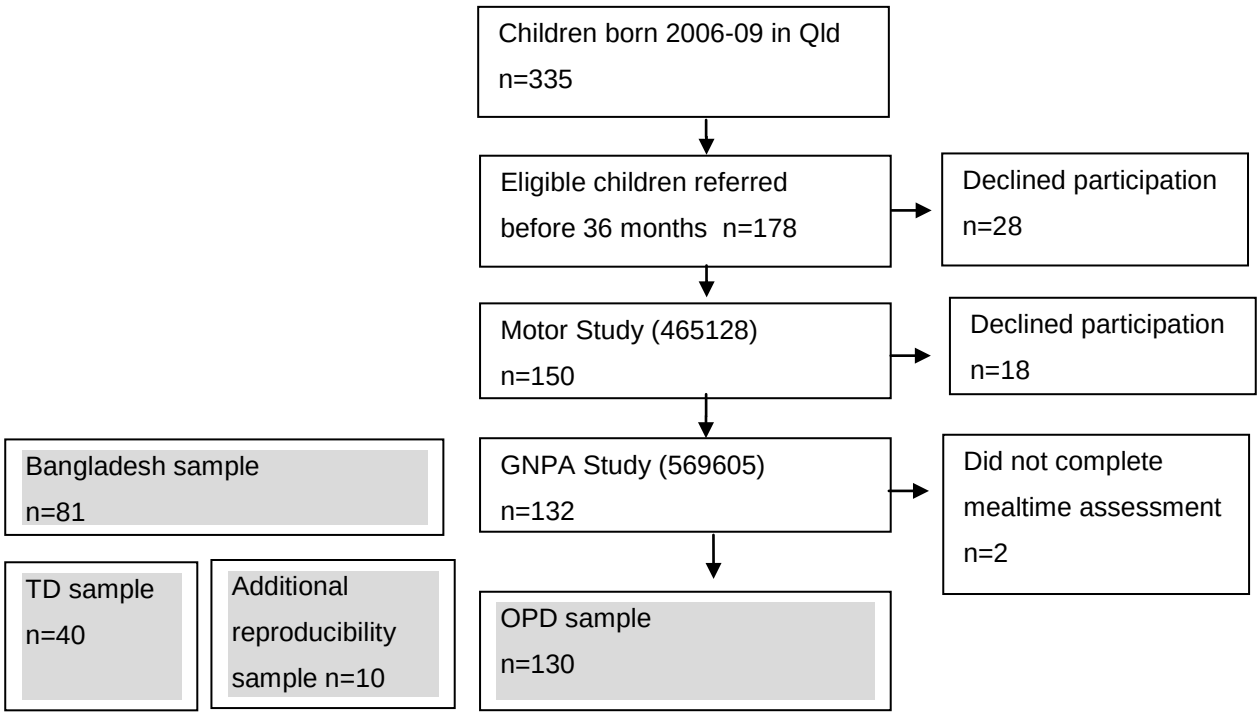

Figure 4. Participant Numbers for Oropharyngeal Dysphagia Study: Growth Nutrition and Physical Activity, Reproducibility and Typically Developing Samples

Abbreviations: CPFQ Queensland Cerebral Palsy Child Feeding Questionnaire; GNPA Growth, Nutrition and Physical Activity, OPD Oropharyngeal Dysphagia; TD Typically Developing

Table 5. Overview of Papers Included in this Thesis, Sample Size and Inclusion Criteria

Paper	n=	Samples included	Representative of CP population	Inclusion criteria for GNPA[a]
Oropharyngeal dysphagia and gross motor skills in children with cerebral palsy	120	GNPA	Gender GMFCS Motor type	Confirmed diagnosis of CP Born in Queensland Birth years 2006-2009 Recruited before August 2012
Validity and reproducibility of measures of oropharyngeal dysphagia in preschool children with cerebral palsy	130 40 40	GNPA TD Reproducibility	Gender GMFCS Motor type	Confirmed diagnosis of CP Born in Queensland Birth years 2006-2009
Oropharyngeal dysphagia in preschool children with cerebral palsy: oral phase impairments	130	GNPA	Gender GMFCS Motor type	Confirmed diagnosis of CP Born in Queensland Birth years 2006-2009
Clinical signs suggestive of pharyngeal dysphagia in preschool children with cerebral palsy	130	GNPA	Gender GMFCS Motor type	Confirmed diagnosis of CP Born in Queensland Birth years 2006-2009
Food and fluid texture consumption in a population-based cohort of preschool children with cerebral palsy: relationship to dietary intake	99	GNPA	Gender	Confirmed diagnosis of CP Born in Queensland Birth years 2006-2009 Complete 3-day food record
Longitudinal study of oropharyngeal dysphagia in preschool children with cerebral palsy	53	GNPA	Gender GMFCS (Ax1) Motor type	Confirmed diagnosis of CP Born in Queensland Birth years 2006-2009 2 assessments before 36 mths
Motor severity in children with cerebral palsy studied in a high-resource and low-resource country	81 223	OPD-Bd CP Child	Gender GMFCS Motor type	NA
Oropharyngeal dysphagia in children with cerebral palsy studied in a high and low resource country	81 130	OPD-Bd GNPA	Gender GMFCS Motor type	Confirmed diagnosis of CP Born in Queensland Birth years 2006-2009

Abbreviations: CP, Cerebral Palsy; CP Child, Brain Structure and Motor Development Study (NHMRC 465128); GNPA, Growth Nutritional and Physical Activity (NHMRC 569605); KAB, Katherine Benfer (candidate); NA, Not Applicable; TD, Children with Typical Development

[a] Inclusion criteria for GNPA sample explains source of difference in study numbers between published papers

Summary of Chapter 3

The literature review described in this protocol identified that there is a paucity of data describing the prevalence of OPD across the full range of GMFCS levels in preschool children with CP. Thus, the first study described in the protocol was undertaken, to describe the overall prevalence of OPD (and its subtypes: oral phase impairments, pharyngeal phase impairments and impaired saliva control) in children with CP aged 18 to 36 months, and its relationship to gross motor function.

Chapter 4: Results – Overall Prevalence of Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy

Introduction to Chapter 4

This chapter presents the published article "Oropharyngeal Dysphagia and Gross Motor Skills in Children with Cerebral Palsy". It describes the overall prevalence of OPD and its subtypes (oral phase impairment, pharyngeal phase impairment, and impaired saliva control) and how this varies according to gross motor function on the GMFCS (with n=120 participants). As described in the previous chapter, there has been a large amount of variability in OPD prevalence estimates reported in the literature, and in part this is related to the heterogeneity of the diagnosis of CP. By considering prevalence by GMFCS level, the results can be more readily compared to other studies and extrapolated to any population of children with CP with similar sample characteristics.

Paper 3: Oropharyngeal Dysphagia and Gross Motor Skills in Children with Cerebral Palsy

This paper was published in Pediatrics and has been cited 18 times (journal impact factor 5.297). This paper was in the year's top 20 articles on developmental disabilities for the American Academy of Cerebral Palsy and Developmental Disability (2013-2014).

> Benfer KA, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Oropharyngeal Dysphagia and Gross Motor Skills in Children with Cerebral Palsy. *Pediatrics*;131(5):e1553-1563.

This paper was also presented as a poster at the 6th Biennial Conference of the Australasian Academy of Cerebral Palsy and Developmental Medicine, 30 May-1 June 2012, Brisbane, Australia.

Benfer KA, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Subtypes of oral motor dysfunction in feeding and its relationship with gross motor skills in young children with cerebral palsy. *Dev Med Child Neurol*. 2012;54(Supp 5):21. (Abstract)

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Oropharyngeal Dysphagia and Gross Motor Skills in Children With Cerebral Palsy

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Pediatrics; originally published online April 15, 2013;
DOI: 10.1542/peds.2012-3093

Oropharyngeal Dysphagia and Gross Motor Skills in Children With Cerebral Palsy

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KEY WORDS
deglutition disorders, dysphagia, feeding, cerebral palsy, prevalence

ABBREVIATIONS
CP—cerebral palsy
DDS—Dysphagia Disorders Survey
GMFCS—Gross Motor Function Classification System
GMFM—Gross Motor Function Measure
MACS—Manual Ability Classification System
OPD—oropharyngeal dysphagia
SOMA—Schedule for Oral Motor Assessment

Ms Benfer and Ms Weir were responsible for acquisition of data; Ms Benfer was responsible for analysis and interpretation of data; Prof Boyd, Ms Benfer, and Ms Weir drafted the manuscript; Profs Davies, Boyd, Drs Bell, and Ware and Ms Weir were responsible for study design and grant writing; Ms Weir and Prof Boyd were responsible for study supervision; and all authors critically reviewed and approved the final manuscript.

This trial has been registered with the ANZTR Register (Trial Registration Number: ACTRN12611000616976).

www.pediatrics.org/cgi/doi/10.1542/peds.2012-3093

doi:10.1542/peds.2012-3093

Accepted for publication Jan 17, 2013

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PEDIATRICS (ISSN Numbers: Print, 0031-4005; Online, 1098-4275).

FINANCIAL DISCLOSURE: *The authors have indicated they have no financial relationships relevant to this article to disclose.*

FUNDING: This project was supported by the National Health and Medical Research Council Postgraduate Medical and Dental Scholarship 1018264 to Ms Benfer, Career Development Fellowship APP1037220 to Prof Boyd, and Project Grants 569605 and 465128.

WHAT'S KNOWN ON THIS SUBJECT: Oropharyngeal dysphagia (OPD) prevalence is 19-99%. OPD based on parent-report is associated with gross motor skills in children with cerebral palsy (CP), however this underestimates prevalence. Almost all children with severe CP have dysphagia; little is known about mild CP.

WHAT THIS STUDY ADDS: The prevalence of directly assessed OPD in preschool children with CP is 85% (70% in GMFCS I; 100% in GMFCS V). OPD was prevalent even in mild CP. Gross motor functional capacity is strongly related to dysphagia severity and prevalence.

abstract

OBJECTIVES: To determine the prevalence of oropharyngeal dysphagia (OPD) and its subtypes (oral phase, pharyngeal phase, saliva control), and their relationship to gross motor functional skills in preschool children with cerebral palsy (CP). It was hypothesized that OPD would be present across all gross motor severity levels, and children with more severe gross motor function would have increased prevalence and severity of OPD.

METHODS: Children with a confirmed diagnosis of CP, 18 to 36 months corrected age, born in Queensland between 2006 and 2009, participated. Children with neurodegenerative conditions were excluded. This was a cross-sectional population-based study. Children were assessed by using 2 direct OPD measures (Schedule for Oral Motor Assessment; Dysphagia Disorders Survey), and observations of signs suggestive of pharyngeal phase impairment and impaired saliva control. Gross motor skills were described by using the Gross Motor Function Measure, Gross Motor Function Classification System (GMFCS), Manual Ability Classification System, and motor type/ distribution.

RESULTS: OPD was prevalent in 85% of children with CP, and there was a stepwise relationship between OPD and GMFCS level. There was a significant increase in odds of having OPD, or a subtype, for children who were nonambulant (GMFCS V) compared with those who were ambulant (GMFCS I) (odds ratio = 17.9, P = .036).

CONCLUSIONS: OPD was present across all levels of gross motor severity using direct assessments. This highlights the need for proactive screening of all young children with CP, even those with mild impairments, to improve growth and nutritional outcomes and respiratory health. *Pediatrics* 2013;131:e1553–e1562

Oropharyngeal dysphagia (OPD) is reported to be prevalent in 19% to 99% of children with cerebral palsy (CP),[1,2] and may lead to inadequate food/ fluid intake and reduced mealtime safety. It is associated with prolonged mealtimes, poor growth and nutritional status, and potential respiratory consequences, which are a major cause of premature mortality.[3,4] This study defines OPD as impairment to any component of the oral-preparatory, oral (propulsive), and/or pharyngeal phases of the swallow, associated with eating, drinking, or controlling saliva.[5] The neurologic lesion that affects an individual's oropharyngeal sensorimotor skills may also influence their gross motor skills, although the extent and severity may vary.[6] An individual's gross motor skills may also influence the maintenance of a stable feeding posture, which can affect eating and swallowing by altering the position and alignment of the oropharyngeal structures and restricting their mobility.[7,8]

There is generally agreement that OPD is positively associated with the severity of gross motor impairment.[1,2,8–11] Assessment of OPD may be conducted directly, using clinical and/ or instrumental evaluation (such as videofluoroscopy), or indirectly through parent report or chart reviews. To date, studies tended to base the estimates of OPD prevalence on indirect measures, and have mostly focused on school-aged children and those with more severe gross motor impairments.[2,8–10,12] This limits our understanding of the prevalence and nature of OPD and its relationship with gross motor skills, particularly in young children and including those with mild gross motor severities. An enhanced understanding of OPD in this subpopulation and its relationship with gross motor skills is important to facilitate early screening and identification of children at risk for poor growth, nutrition, and respiratory health. The aim of this study was to determine the prevalence of OPD and its subtypes (oral phase, pharyngeal phase, and saliva control) using direct clinical assessment of feeding with standardized measures of OPD, and to investigate the association between gross motor functional skills and OPD. It was hypothesized that OPD would be present across gross motor severity levels, and increase in prevalence and severity as gross motor severity increased.

METHODS

This is a cross-sectional population-based study of preschool-aged children with CP, conducted in Queensland, Australia, between April 2009 and August 2012. It is part of a longitudinal study exploring the relationship among growth, nutrition, and physical activity (Queensland CP Child: Growth, Nutrition and Physical Activity, National Health and Medical Research Council 56960). The design of the larger study[13] and current study[14] have been described elsewhere. Ethics approval was gained through the University of Queensland Medical Research Ethics Committee (2008002260), the Children's Health Services District Ethics Committee (HREC/08/QRCH/112), and other regional and organizational ethics committees (see protocol papers for full list). All families gave written informed consent to participate.

Patients

Children with a confirmed diagnosis of CP, 18 to 36 months corrected age at the time of initial assessment, and born in Queensland between 2006 and 2009, were invited to participate in the study. Children with neurodegenerative conditions were excluded from the study.

Measures

Measures of OPD were selected after conducting a comprehensive systematic review of the psychometric properties and clinical utility.[14,15] Included measures were the following:

1. Schedule for Oral Motor Assessment (SOMA): a discriminative measure that identifies oral motor dysfunction in children according to skills that are typically mastered from 8 to 24 months.[16] It categorizes oral motor dysfunction based on cut-scores for 7 oral motor challenge categories (puree, semisolid, solid, cracker, bottle, trainer cup, cup).[16] The SOMA is predominantly a test of oral phase dysfunction; however, some items pertain to the pharyngeal phase. The assessment of feeding position (upright with/without back support, upright with trunk support, semi-sitting, and supine) was used as a covariate in models.
2. Dysphagia Disorders Survey–Pediatric (Part 2) (DDS): an evaluative measure for screening signs of oral, pharyngeal, and esophageal phase dysphagia in children and adults with a developmental disability.[17] Part 2 provides a raw score that indicates an individual's functional eating competency (maximum impairment raw score of 22) and this subtest has been used previously as a measure of OPD.[2]
3. Clinical signs suggestive of pharyngeal phase impairment: a determination of pharyngeal phase impairment was noted if the child demonstrated any 1 of 16 signs, rated live, and from video by the speech pathologist (see Appendix).
4. Thomas-Stonell Greenberg Saliva Severity Scale: a semiquantitative assessment of drooling severity (1- to 5-point scale of no drooling to profuse drooling) based on observations of anterior saliva loss.[18]

Functional gross motor skills were directly evaluated using the Gross Motor

Function Measure-88 (GMFM-88) for domain scores, and the Rasch-analyzed GMFM-66.[19] From this, children were classified on the Gross Motor Function Classification System (GMFCS) according to their age by using the <2 years and 2- to 4-year scales.[20] The Manual Ability Classification Scale (MACS) was used to classify children's functional upper limb skills.[21] The type of CP (spastic, dyskinetic, hypotonic/ataxic) and motor distribution (hemiplegia, diplegia, quadriplegia) were classified according to the Surveillance of CP in Europe.[22] Children's ability to sit on a mat and maintain head upright for 10 seconds, and sit on a bench for 10 seconds with feet supported were used to indicate head and trunk instability, respectively.

Procedures

Children attended the hospital for mealtime and gross motor assessment. Mealtimes were videoed as recommended in the SOMA administration manual, with children well positioned in their typical mealtime seating. Three standardized presentations of 4 textures (puree, lumpy, chewable, and fluid) were presented by the primary carer, using their regular utensils.[16] Following these standard presentations, the child was allowed to complete the snack as usual. A trained researcher recorded 4 signs suggestive of pharyngeal phase impairment, and severity of drooling before and after the mealtime. All gross motor ratings were conducted by 2 trained physiotherapists.

Reproducibility Study

Twenty children (4 from each GMFCS level) were selected randomly by an independent researcher for analysis of intrarater and interrater reproducibility of all OPD measures and interrater reproducibility for MACS. The clinicians rating the videos were blinded to reliability case status. Intrarater reliability ratings were performed 2 weeks after initial ratings. Interrater reliability ratings were completed independently by 2 speech pathologists for the OPD measures (K.A.B., K.A.W.), and 2 physiotherapists for MACS.

Statistical Analysis

Demographic data were presented with descriptive statistics, and sample representativeness determined in relation to an Australian register study[23] using χ^2 test for trend. Inter- and intrarater reproducibility were assessed by using Cohen's κ (unweighted and weighted) and percentage agreement. The association between gross motor skill attainment and OPD were analyzed by using the χ^2 test for trend, and individual motor categories compared using logistic regression. Univariate

TABLE 1 Characteristics of Participants in the OPD Study

	Participants, *n* (%)	Australian Register Study	*P* Value[a]
Birth year		n/a	
2006	17 (14.2)		
2007	41(34.2)		
2008	34 (28.3)		
2009	28 (23.3)		
Gender, male	74 (61.7)	n/a	
GMFCS level			.220
I	50 (41.7)	114 (35.0)	
II	17 (14.2)	53 (16.0)	
III	22 (18.3)	46 (14.0)	
IV	11 (9.2)	52 (16.0)	
V	20 (16.7)	58 (18.0)	
Primary motor type			.087
Spasticity	104 (86.7)	279 (86.4)	
Dyskinesia	6 (5.0)	5 (2.0)	
Ataxia	1 (0.8)	9 (3.0)	
Hypotonia	9 (7.5)	9 (3.0)	
Motor distribution			
Unilateral	36 (30.0)	98 (30.3)	.913
Diplegia	31 (25.8)	78 (24.0)	
Triplegia/Quadriplegia	53 (44.2)	147 (45.7)	
Preterm birth (<37 wk)	62 (51.7)	n/a	
Tube feeding (partial or total)	13 (10.8)	n/a	
Geographical location		n/a	
Highly accessible	80 (66.7)		
Moderately accessible	16 (13.3)		
Accessible	21 (17.5)		
Remote	3 (2.5)		
Very remote	0 (0.0)		

n/a, data not available.
[a] *P* value for χ^2 test for trends.

TABLE 2 Reproducibility of OPD Measures

	Interrater		Intrarater	
	Reliability[a]	% Agreement	Reliability[a]	% Agreement
Overall OPD	κ 1.00	100.00	κ 0.77	95.00
SOMA (overall)	κ 0.90	95.00	κ 1.00	100.00
DDS-Part 2 (overall)	κ 1.00	100.00	κ 0.86	95.00
DDS-Part 2 (raw score)	ICC 0.99	72.22[b]	ICC 0.99	50.00[b]
Pharyngeal signs and symptoms (overall)	κ 0.77	90.00	κ 0.88	95.00
Impaired saliva control (overall)	κ 0.89	94.74	κ 1.00	100.00

ICC, intraclass correlation coefficient; κ, Cohen's κ coefficient.
[a] $P < .001$. κ <0 poor, 0.01–0.20 slight, 0.21–0.40 fair, 0.41–0.60 moderate, 0.61–0.80 substantial, and 0.81–1.0 almost perfect.[31]
[b] Lower agreement for DDS-Part 2 raw score, as this is an interval scale (0–22).

logistic regression analyses were undertaken for all explanatory variables of interest (age, gender, geographical accessibility, preterm status, postural instability, and supported feeding position). Variables consistently significant at the $P = .05$ level were then included in all multivariate regressions. All data analyses were performed by using Stata (Stata Corp, College Station, TX).

RESULTS

Sample Characteristics

There were 166 eligible children referred, of which 122 parents consented to participate in the Growth, Nutrition and Physical Activity study, and 120 children completed the requirements to participate in the OPD study. Of the children who declined participation, 18 participated in only the concurrent CP Child Motor Study,[13,14] and 26 declined both studies (8 because of study burden, 12 because of family circumstances, 2 were non-English speaking, 3 resided interstate, and 1 died). Participants' ages ranged from 17 to 37 months corrected age at the time of assessment (mean = 27.0 months, SD = 5.2). Partial or total tube feeding was present in 10.8% of the sample at the time of assessment. Characteristics of the sample are presented in Table 1.

Reproducibility of Measures

The results from the OPD reproducibility study are presented in Table 2. The inter- and intrarater agreement were >90% for all binary OPD measures, and reliability was substantial to perfect ($P < .001$). There was a strong correlation between DDS Part 2 scores for intrarater (intraclass correlation coefficient = 0.99, $P < .001$) and interrater (intraclass correlation coefficient = 0.99 $P < .001$). The MACS had 60% perfect agreement, and 36% near perfect agreement (within 1 level), with moderate reliability (weighted $\kappa = 0.47$, $P < .001$).

Prevalence of OPD and Its Relationship With Motor Function

Overall, 85% of children had OPD identified on 1 or more direct clinical measures (SOMA, DDS, or pharyngeal signs), excluding impaired saliva control, which was considered developmentally appropriate (Table 3). There was a significant increasing trend for the prevalence of all OPD variables as GMFCS level increased ($P < .05$) (Fig 1).

The relationships between GMFCS and OPD are presented in Table 3. Postural instability and position were consistently significantly associated with the OPD outcomes in all univariate models (largest P value for instability = .008, and for position was .052). No other explanatory variables were significant for any outcome (with the exception of geographical access for pharyngeal signs, $P = .009$); therefore, instability and position were included in all

TABLE 3 Relationship Between GMFCS and OPD

	n (%)	Crude Odds Ratio (95% CI)	*P* Value	Adjusted Odds Ratio[a] (95% CI)	*P* Value
OPD overall	102 (85.0)	—	—	—	—
I	35 (70.0)	1.0 (reference)	—	1.0 (reference)	—
II	14 (82.4)	2.0 (0.5–8.0)	.327	1.8 (0.4–7.6)	.403
III	22 (100.0)	19.7 (1.2–403.1)[b]	.024	∞ (n/c)	n/c
IV	11 (100.0)	10.0 (1.0–∞)[b]	.052	∞ (n/c)	n/c
V	20 (100.0)	17.9 (1.1–368.0)[b]	.036	∞ (n/c)	n/c
Postural instability	31 (30.4)	16.3 (1.8–∞)[b]	.005	∞ (n/c)	n/c
Supported feeding position	38 (40.0)	24.8 (1.7–504.0)[b]	.004	∞ (n/c)	n/c
SOMA overall	51 (42.5)	—	—	—	—
I	9 (18.0)	1.0 (reference)	—	1.0 (reference)	—
II	5 (29.4)	1.9 (0.5–6.8)	.322	1.6 (0.4–5.8)	.511
III	9 (40.9)	3.2 (1.0–9.6)	.043	2.0 (0.6–6.8)	.282
IV	8 (72.7)	12.2 (2.7–55.0)	.001	4.6 (0.7–28.5)	.104
V	20 (100.0)	171.9 (9.9–3476.9)[b]	.000	∞ (n/c)	n/c
Postural instability	27 (52.9)	12.4 (4.1–37.3)	.000	2.7 (0.6–11.8)	.197
Supported feeding position	28 (62.2)	6.1 (2.9–12.8)	.000	1.7 (0.7–4.5)	.278
DDS overall	94 (78.3)	—	—	—	—
I	28 (56.0)	1.0 (reference)	—	1.0 (reference)	—
II	14 (82.4)	3.7 (0.94–14.4)	.062	3.5 (0.9–13.9)	.082
III	21 (95.5)	16.5 (2.1–132.4)	.008	19.0 (1.9–193.8)	.013
IV	11 (100.0)	18.2 (2.0–∞)[b]	.004	∞ (n/c)	n/c
V	20 (100.0)	32.4 (2.2–709.9)[b]	.001	∞ (n/c)	n/c
Postural instability	31 (33.0)	26.3 (3.0–∞)[b]	.000	∞ (n/c)	n/c
Supported feeding position	36 (41.4)	4.2 (1.4–13.0)	.013	0.4 (0.8–2.2)	.305
Pharyngeal phase impairment	74 (61.7)	—	—	—	
I	23 (46.0)	1.0 (reference)	—	1.0 (reference)	
II	7 (41.2)	0.8 (0.3–2.5)	.730	0.7 (0.2–2.3)	.531
III	16 (71.4)	3.1 (1.1–9.3)	.040	1.9 (0.6–6.3)	.270
IV	9 (81.8)	5.3 (1.0–27.0)	.045	0.8 (0.1–7.5)	.852
V	19 (95.0)	22.3 (2.8–179.7)	.004	0.3 (0.0–10.6)	.506
Postural instability	29 (39.2)	10.0 (2.4–41.4)	.001	4.5 (0.8–23.6)	.079
Supported feeding position	34 (50.8)	6.0 (2.3–15.3)	.000	4.3 (1.1–17.3)	.042
Impaired saliva control	54 (47.4)	—	—	—	—
I	18 (36.0)	1.0 (reference)	—	1.0 (reference)	—
II	6 (35.3)	1.0 (0.3–3.1)	.958	0.9 (0.3–2.9)	.844
III	12 (54.5)	2.1 (0.8–5.9)	.145	2.0 (0.7–5.8)	.234
IV	8 (72.7)	4.7 (1.1–20.2)	.035	4.4 (0.7–26.4)	.104
V	10 (71.4)	4.4 (1.2–16.2)	.024	2.5 (0.3–22.0)	.423
Postural instability	18 (33.3)	3.0 (1.3–6.6)	.008	2.2 (0.8–6.6)	.148
Supported feeding position	24 (44.4)	1.7 (1.0–2.9)	.052	0.8 (0.4–1.8)	.585

CI, confidence interval; n/c, not calculable; —, no data available.

[a] Adjusted odds ratios for perfectly predicted are reported as ∞ (95% CI n/c).

[b] Exposure predicts outcome perfectly, therefore calculated in episheet, based on Fisher's Exact Test.

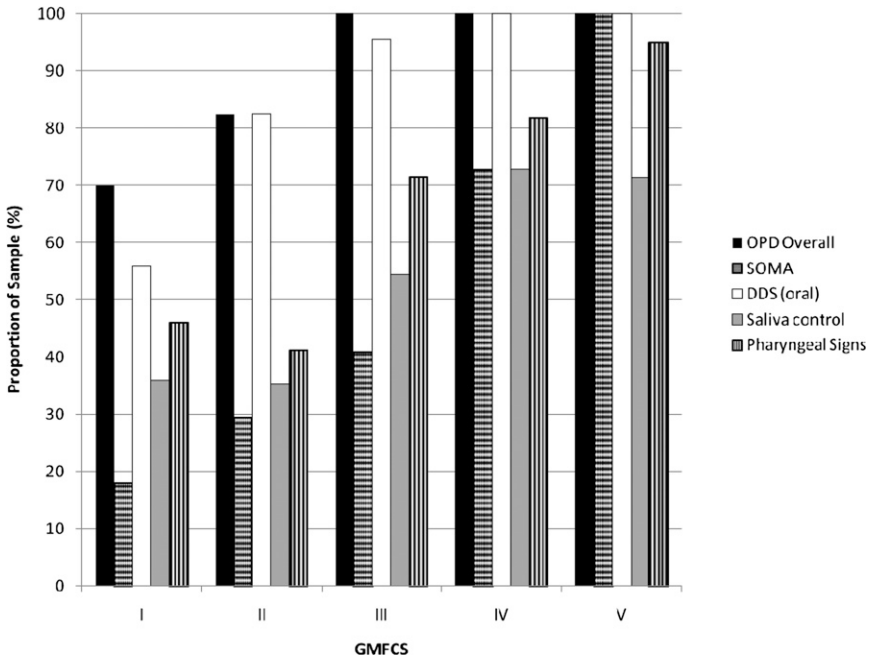

FIGURE 1

Proportion of oropharyngeal dysphagia by subtype, according to GMFCS. Key: OPD $P < .001$; SOMA $P < .001$; DDS $P < .001$; saliva control $P = .002$; pharyngeal signs $P < .001$.

multivariate models. For the overall OPD model, postural instability and supported feeding position perfectly predicted the presence of OPD (that is, no children with head or trunk instability, or fed in a supported feeding position had typical oral feeding skills); therefore, adjusted odds ratios could not be reported. The relationship between OPD and MACS is presented in Table 4. The results showed a similar finding to GMFCS models, with children classified in the severe levels for manual ability having significantly higher odds of OPD.

The relationship between OPD prevalence and gross motor capacity overall and by motor domain is reported in Table 5. For each unit increase in GMFM score, the odds of having OPD increased by 2% to 11% (odds ratio = 0.98 and 0.89, respectively). The severity of OPD, based on the DDS Part 2 raw score, was significantly correlated with motor severity on the GMFM-66 (Fig 2).

The relationship between motor type/distribution and prevalence of OPD are presented in Table 6. All children with 4-limb involvement had OPD. Of the children with diplegia (spastic) and OPD, GMFM-66 scores were significantly lower than those with diplegia and no dysphagia (GMFM = 53.9, SD = 8.2 for OPD, compared with GMFM = 60.6, SD = 4.6 for no OPD [$P = .028$]). A similar but not statistically significant trend was noted in the unilateral spasticity group, of a GMFM score of 57.4, SD = 7.3, for OPD compared with 61.9, SD = 11.34, for those without OPD ($P = .174$).

DISCUSSION

This population-based study found 85% of children with CP aged 18 to 36 months had OPD, based on impairment on 1 or more of the SOMA, DDS, or pharyngeal signs. Estimates for the subtypes varied markedly from 43% on the SOMA, to 78% on the DDS, both of which are primarily measures of oral phase impairment. Pharyngeal phase impairment and impaired saliva control, identified through standardized clinical observations, were present in about half of the sample (62% and 48%, respectively).

The overall OPD estimate is consistent with the prevalence estimates reported in the only 2 previous studies conducted in preschool children with CP of 78%[24] and 90%.[11] However, the estimate by Reilly et al[11] of 90%, obtained through direct assessments of a community-based sample, used the SOMA alone, which is a significantly higher estimate than ours of 40% found using only the SOMA. This discrepancy likely reflects the bias toward recruitment of participants with more severe gross motor impairments in the study by Reilly et al[11] (70% had severe-profound motor impairment), when in fact the distribution of gross motor severity tends to be skewed to the milder end of the range.[23] The estimate in the study by Wilson and Hustad[24] was based on clinical evidence of oral-motor involvement, which was a broad classification of any neurologically based impairment of speech subsystems. Tube feeding or cough/choke/gag were identified by parent report in close to all of these children (73%). The gross motor severity of children in this study sample was not reported, thus their estimate may also reflect a bias toward recruitment of children with more severe gross motor severities. The current study estimate strengthens previous estimates by using direct clinical OPD measures with strong reproducibility and a representative study sample across gross motor severity levels.

There were an increasing number of children with OPD for each increase in GMFCS level, and this difference between groups was statistically significant for each subtype. OPD was present across all gross motor severity levels, with as few as 18% of children in GMFCS I identified as having OPD using the SOMA, and as many as 56% of children in this group using the DDS. Although the trend for increasing prevalence of OPD with increased gross motor severity

TABLE 4 Relationship Between Manual Ability and OPD

	n (%)	Odds Ratio (95% CI)	*P* value	Adjusted Odds Ratio[a] (95% CI)	*P* value
OPD overall	—	—	—		
I	31 (70.5)	1.0 (reference)	—	1.0 (reference)	
II	39 (88.6)	3.3 (1.1–10.2)	.041	2.6 (0.8–8.3)	.107
III	5 (100.0)	4.7 (0.4–∞[b]	.307	∞ (n/c)	n/c
IV	9 (100.0)	8.1 (0.8–∞)[b]	.101	∞ (n/c)	n/c
V	18 (100.0)	15.9 (0.9–327.7)[b]	.065	∞ (n/c)	n/c
SOMA overall	—	—	—	—	—
I	7 (15.9)	1.0 (reference)	—	1.0 (reference)	
II	13 (29.6)	2.2 (0.8–6.2)	.132	1.9 (0.7–5.7)	.232
III	4 (80.0)	21.1 (2.1–218.5)	.010	14.8 (1.1–197.6)	.042
IV	9 (100.0)	95.0 (7.8–∞)[b]	.000	∞ (n/c)	n/c
V	18 (100.0)	185.0 (9.5–3556.6)[b]	.000	∞ (n/c)	n/c
DDS overall	—	—	—		
I	28 (63.6)	1.0 (reference)	—	1.0 (reference)	
II	34 (77.3)	1.9 (0.8–5.0)	.164	1.8 (0.7–4.8)	.235
III	5 (100.0)	6.4 (0.6–∞)[b]	.149	∞ (n/c)	n/c
IV	9 (100.0)	11.0 (1.2–∞)[b]	.032	∞ (n/c)	n/c
V	18 (100.0)	21.4 (1.4–481.6)[b]	.013	∞ (n/c)	n/c
Pharyngeal phase impairment	—	—	—		
I	18 (40.9)	1.0 (reference)	—	1.0 (reference)	
II	26 (59.1)	2.1 (0.9–4.9)	.090	1.7 (0.7–4.2)	.237
III	4 (80.0)	5.8 (0.6–56.1)	.130	1.9 (0.1–25.8)	.627
IV	8 (88.9)	11.6 (1.3–100.6)	.027	1.7 (0.1–28.4)	.718
V	18 (100.0)	53.0 (3.5–1165.4)[b]	.000	∞ (n/c)	n/c
Impaired saliva control	—	—	—		
I	15 (34.1)	1.0 (reference)	—	1.0 (reference)	
II	18 (40.9)	1.3 (0.6–3.2)	.509	1.5 (0.6–3.6)	.410
III	6 (100.0)	20.9 (1.8–∞)[b]	.008	∞ (n/c)	n/c
IV	7 (88.9)	15.5 (1.8–135.5)	.013	33.5 (1.8–610.0)	.018
V	8 (66.7)	3.9 (1.0–15.0)	.050	4.2 (0.3–54.1)	.266

CI, confidence interval; n/c, not calculable.

[a] Multivariate models include postural instability and position. These covariates did not reach statistical significance for any outcome. Odds ratios for perfectly predicted are reported as ∞ (95% CI n/c).

[b] Exposure predicts outcome perfectly, therefore calculated in episheet, based on Fisher's Exact Test.

was stepwise for each GMFCS level, these relationships were generally only significant for children in GMFCS III to V compared with GMFCS I. All children who were tube fed were from GMFCS IV to V.

The proportion of children with OPD from the more severe gross motor groups was consistent with other studies. Direct ratings of children's mealtimes were conducted in the studies by Calis et al[2] and Santoro et al,[25] finding OPD in almost all children (99% and 100% respectively) from GMFCS IV to V, which was also found in the current study. In a large register-based study of children (median age 5 years) (n = 1357), there was a 5-fold increase in odds for GMFCS IV and a 15-fold increase for GMFCS V of having swallowing/chewing difficulties and excessive drooling.[1] This increase in likelihood with GMFCS is comparable to the magnitude found in the current study, although the prevalence of OPD overall and by GMFCS level was markedly higher in the current study by using direct assessments. Using validated measures (SOMA and Standard Recording of Central Motor Deficit), the presence of gross motor impairment was significantly associated with the presence of oral motor dysfunction in a cross-sectional community-based sample of 49 preschool children with CP.[11] Although strengthened by using validated measures for both oral motor and gross motor skills, the sample was small, skewed to more severe gross motor severity levels, and only binary variables were used (presence/absence of dysfunction).

Children with mild gross motor impairments have received limited attention in the literature to date. A study of ambulatory (with or without assistive mobility) school-aged children (estimated to be GMFCS I to III) with mild CP performed more poorly than controls on spoon feeding, biting, and cup drinking using direct clinical assessment on the Functional Feeding Assessment modified.[26] The specific prevalence of feeding difficulties or influence of gross motor skill could not be ascertained from the data. Another study investigating parent-reported feeding difficulties in children using GMFCS to classify gross motor level identified just 4% of children from GMFCS I to III with feeding difficulties, compared with 22% of children from GMFCS IV to V.[27] The prevalence of OPD in the mild motor groups found in the current study are greater than previously documented, and may indicate the underdetection of mild feeding difficulties, particularly when using indirect assessments. The background prevalence in typically developing children and the potential effects of mild OPD on health warrant further investigation.

The results showed that the higher the gross motor capacity score on the GMFM, the fewer children had OPD (2% to 11% reduced chance of OPD with each increase in GMFM-66 score). This was statistically significant overall and for the 30- to 36-month stratum, but not for the 18- to 24-month group. Eighteen to 24 months is a period of significant gross motor maturation, which could explain the insignificant association in this age range. Almost three-quarters of the variability seen in the severity of OPD (by DDS part 2 raw score) could be explained by gross motor functional capacity. The gross motor domain with the greatest association with OPD

prevalence was the sitting domain, which is consistent with the literature, that suggests postural stability and trunk control are important for feeding success.[2,8] This was further supported by the influence of postural instability (head and trunk) and feeding position on the GMFCS model.

TABLE 5 Relationship Between Motor Capacity on the GMFM and Prevalence of OPD

	Odds Ratio[a]	95% Confidence Interval	*P* Value
GMFM-66 overall[b]	0.89	0.84–0.95	.001
18–24 mo	0.90	0.81–1.00	.054
30–36 mo[b]	0.93	0.88–0.99	.017
GMFM-88 (A) lying, rolling[b]	0.96	0.92–0.99	.020
18–24 mo	0.95	0.89–1.01	.126
30–36 mo	0.97	0.93–1.01	.121
GMFM-88 (B) sitting[b]	0.92	0.87–0.97	.004
18–24 mo	0.95	0.89–1.01	.078
30–36 mo[b]	0.90	0.82–0.98	.017
GMFM-88 (C) crawling, kneeling[b]	0.96	0.94–0.99	.002
18–24 mo	0.98	0.95–1.01	.118
30–36 mo[b]	0.95	0.91–0.99	.015
GMFM-88 (D) standing[b]	0.96	0.94–0.98	.001
18–24 mo	0.97	0.94–1.00	.050
30–36 mo[b]	0.95	0.92–0.99	.013
GMFM-88 (E) walking, running, jumping[b]	0.96	0.94–0.98	.000
18–24 mo	0.95	0.92–1.00	.027
30–36 mo[b]	0.96	0.93–0.99	.007

[a] Crude odds ratios reported as covariates of postural instability and position predict perfectly for outcome.
[b] Statistically significant.

The number of children with OPD was strongly linked to motor type and distribution, with distributions affecting 3 or more limbs (spastic quadriplegia, dyskinesias, and hypotonia/ ataxia) resulting consistently in OPD. As dyskinesias and hypotonia are less common in the CP population, the small numbers meant that these relationships were not statistically significant, despite OPD being consistently present. This finding is consistent with the Oxford Feeding Study[9] of 271 school-aged children with OPD, which found that those with more extensive motor involvement (ie, quadriplegia and dyskinesia) are most likely to have difficulties with swallowing and articulation. In our study, about 70% of children with hemiplegia or diplegia had OPD, and these children had lower average scores on the GMFM than children with hemiplegia/digplegia and no feeding difficulties. This suggests that motor type/distribution in conjunction with functional severity may be useful in predicting children at risk for feeding difficulties.

This study explored OPD in a large representative sample of young children with CP using direct clinical measures. Although overall the sample size was adequate, the lower occurrence of certain phenomena in children with CP limited the statistical power for some individual analyses. Another limitation in this study was sampling an age range that crossed 2 different GMFCS scales (<2 years and 2–4 years). Children's GMFCS level may be reclassified after their second birthday, which may affect comparisons across the sample. The most significant limitation in all studies of feeding in young children remains the lack of a gold standard or consensus in the definition for the construct of OPD. A large range was found in the identification of OPD cases using each of the OPD measures, particularly between the SOMA and DDS. The SOMA was designed to detect clinically significant OPD, and therefore may lack sensitivity in detecting mild OPD. Conversely, although the DDS and pharyngeal signs appear to be detecting the milder feeding difficulties, these measures may be misclassifying behaviors as OPD that are present in young typically developing children. Although normative data exist for feeding efficiency[28,29] and parent-reported acquisition of a limited number of oral behaviors,[30] our future work assessing a typically developing reference sample with the SOMA and DDS will address some of the questions surrounding the validity of measures. Triangulation of videofluoroscopy swallow study results with clinical pharyngeal signs will further validate these findings, and will be the subject of future articles.

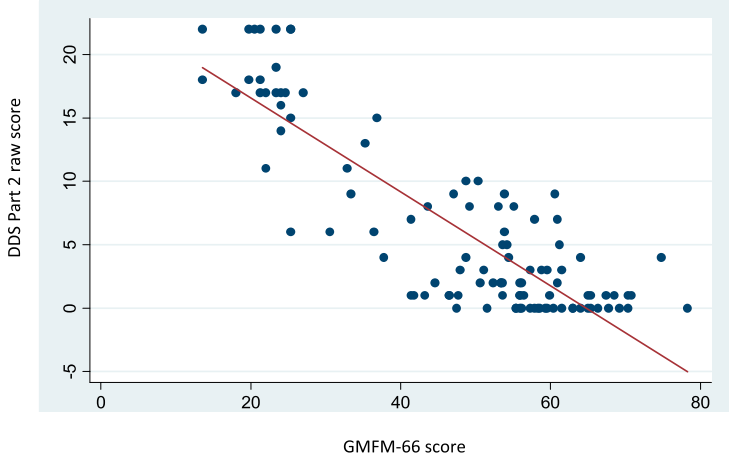

FIGURE 2
Relationship between DDS raw score and GMFM-66. Key: Pearson's correlation: $r = -0.85$, $r^2 = 0.73$, $P < .000$; 18–24-month subgroup $r = -0.85$, $r^2 = 0.72$, $P < .000$; 30–36-month subgroup $r = -0.83$, $r^2 = 0.68$, $P < .000$.

CONCLUSIONS

This study proposes a more plausible OPD estimate of 85% to reflect the prevalence of OPD in young children with CP based on direct clinical measures with reported validity and reliability, and a representative population-based sample across the range of gross

TABLE 6 Relationship Between OPD and Motor Type and Distribution

	OPD Overall, *n* (%)	Odds Ratio (95% Confidence Interval)[a]	*P* Value
Spastic unilateral (base)	27 (75.0)	—	—
Spastic diplegia	22 (71.0)	0.8 (0.3-2.4)	.711
Spastic quadriplegia[b]	37 (100.0)	25.9 (2.4-∞)[c]	.002
Dyskinesia	6 (100.0)	4.5 (0.2-98.4)[c]	.983
Hypotonia/ ataxia	10 (100.0)	7.3 (0.4-154.8)[c]	.525

[a] Crude odds ratios reported as covariates of postural instability and position predict perfectly for outcome.
[b] Classification includes triplegia (ie, bilateral spasticity with >2 limbs involved).
[c] Exposure predicts outcome perfectly, therefore calculated in episheet, based on Fisher's Exact Test.

motor severity levels. This study has confirmed previous findings, that OPD is related to gross motor severity, using a universally recognized gross motor classification (GMFCS). OPD was present across all GMFCS levels, which highlights the need for proactive screening of all young children with CP, even those from GMFCS I, to detect children at risk for feeding-related growth, nutrition, and respiratory compromise. To better understand the nature of OPD in this group of children, the OPD measures need further testing of their psychometric properties, particularly with reference to a typically developing sample. In addition, studies highlighting the specific impairments of children during the oral and pharyngeal phases of the swallow, and longitudinally during maturation of oral sensorimotor skills, will enable clinicians and researchers to better design and target interventions for children with feeding difficulties.

ACKNOWLEDGMENTS

We would like to thank Laura Pareezer (BN), Clinical Trials Coordinator QCPRRC for her time on recruitment and administrative support. We would also like to thank Physiotherapists Rachel Jordan (BPhty) and Chris Finn (BPhty) for data collection and gross motor ratings; and Dietitians Joanne McMah (M Nutr & Diet), Stina Oftedal (B.Hlth.Sc (Hons) Nutr & Diet) and Jacqueline Walker (Bsc) for data collection of feeding videos.

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APPENDIX Signs Suggestive of Pharyngeal Phase Impairment

Gags when eating or drinking.
Coughs when eating or drinking.
Chokes when eating or drinking.
Vomits when eating or drinking.
Clears his/her throat often during or after meals.
Needs to swallow a number of times to clear each mouthful of food or drink.
Wheezes during/after eating or drinking.
Has "stridor" when breathing in or out during eating or drinking.
Becomes breathless and breathes quickly during eating or drinking.
Breathing becomes labored or effortful during eating or drinking.
Has a "rattly chest" after eating or drinking.
Gets a "snuffly nose" after eating or drinking.
Has a "gurgly voice" after eating or drinking.
Has wet or "gurgly" breathing during or after eating or drinking.
Has runny eyes or "eye tearing" after swallows of certain food or drinks.
Seems to go "blue" around the lips/face or turn "dusky" or pale after drinking or eating.
Generally refuses to eat or drink some food or fluid textures.

Oropharyngeal Dysphagia and Gross Motor Skills in Children With Cerebral Palsy

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Pediatrics; originally published online April 15, 2013;
DOI: 10.1542/peds.2012-3093

Summary of Chapter 4

This paper found 85% of preschool children with CP had OPD, defined as impairment on 1 or more measures (including the SOMA, DDS, and clinical signs suggestive of pharyngeal phase impairment). Direct clinical measures were used to assess OPD, and children were sampled from across the full spectrum of gross motor functional severity (in a representative sample). Oropharyngeal dysphagia prevalence increased with poorer gross motor function. This paper contributed to our knowledge of OPD in preschool children with CP in the following areas:

i. OPD was prevalent, even in children with ambulatory CP (GMFCS I-II). This group has received limited attention in the literature to date. Studies have suggested these children perform more poorly on functional feeding tasks than children with TD.[38] Only 2 prevalence estimates were reported previously in the literature for this subgroup, with large discrepancy. One study estimate was 4% based only on parent report,[94] and the other reported 70% impairment using VFSS, although the sample size was small (n=10).[72]

ii. OPD prevalence increased with each increase in GMFCS level, and this was statistically significant. All children from GMFCS III-V had OPD (on 1 or more measures). The most available data in the literature describe children with nonambulatory CP (ie, GMFCS IV-V).[1,62] These 2 studies used direct ratings of OPD, finding it prevalent in almost all children with nonambulatory CP (100% and 99%, respectively). This was consistent with our study findings.

iii. *Postural instability* (defined as inability to sit on a bench for 10 seconds) and *supported feeding position* (mealtime in supportive seating, caregiver's lap) were both significantly related to each of the OPD outcomes (except for *supported feeding position* with OPD on the DDS, and with impaired saliva control).

iv. The prevalence of OPD was not only related to gross motor functional performance (GMFCS), but also to gross motor functional capacity (as measured on the Gross Motor Function Measure [GMFM]). Almost three quarters of the variability seen in OPD severity (DDS raw score) could be attributed to gross motor functional capacity. OPD was most strongly related to a child's ability to sit (ie, the sitting domain on the GMFM).

v. All children with 4-limb involvement (4-limb bilateral spasticity, dystonia, athetosis, hypotonia and ataxia) had OPD. As dykinesias, hypotonia and ataxia are relatively uncommon in the CP population, the small numbers meant that these relationships were not statistically significant, despite OPD being consistently present.The GMFM scores of children with 2-limb involvment with and without OPD also differed markedly.

This suggests gross motor functional severity in conjunction with motor type are important to consider in relation to OPD.

vi. There was a large discrepancy between OPD detected on the DDS (78%) and OPD on the SOMA (42%). Both of these measures appear to assess the same construct (focused on the oral phase, but with some items pertaining to the pharyngeal phase).

Based on the large discrepancy noted in the standardised measures of OPD (SOMA and DDS) in this article, it was decided to conduct a substudy exploring the psychometrics of these measures in children with CP aged 18 to 36 months. This substudy, described in the following chapter, explored the construct validity and reproducibility of the SOMA and DDS, in addition to a third measure, the PSAS.

Chapter 5: Validation of Measures of Oropharyngeal Dysphagia for Preschool Children with Cerebral Palsy

Introduction to Chapter 5

This chapter presents the published article “Validity and Reproducibility of Measures of Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy”. This substudy was prompted by the large discrepancy in estimates yielded by the measures (SOMA and DDS) in the OPD prevalence study reported in the previous chapter. The substudy presented in this chapter, aimed to elucidate the measure which was most accurately detecting OPD in preschool children with CP. One of the greatest challenges of research in OPD is the lack of a gold standard measure. In the absence of a gold standard, the pursuit to determine the best measure was 3-fold: (1) testing the reproducibility (interrater and intrarater) of each of the measures, (2) testing the construct validity of the measures by triangulating the SOMA, DDS and PSAS; as well as using a latent-class model (using web-based software) to estimate prevalence in the absence of a gold standard, (3) testing the discriminative validity by applying the 3 measures to a sample of children with TD. This paper forms a critical foundation for the papers which follow this chapter, in order to better understand the measures’ psychometric properties.

Paper 4: Validity and Reproducibility of Measures of Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy

This article was published in Developmental Medicine and Child Neurology, and has been cited once (journal impact factor 3.292). Reproduced from Developmental Medicine and Child Neurology, Published online 7 November 2014 DOI: 10.1111/dmcn.12616 , Benfer, KA, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN, Validity and reproducibility of measures of oropharyngeal dysphagia in Preschool Children with Cerebral Palsy, pages no., Copyright 2014, © 2014 Mac Keith Press, with permission from John Wiley and Sons.

Benfer KA, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Validity and reproducibility of measures of oropharyngeal dysphagia in preschool children with cerebral palsy. *Dev. Med. Child Neurol.* 2014. http://dx.doi.org/10.1111/dmcn.12616. Accessed November 16, 2014.

Validity and reproducibility of measures of oropharyngeal dysphagia in preschool children with cerebral palsy

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PUBLICATION DATA
Accepted for publication 6th September 2014.
Published online

ABBREVIATIONS

DDS	Dysphagia Disorders Survey
OPD	Oropharyngeal dysphagia
PSAS	Pre-Speech Assessment Scale
SOMA	Schedule for Oral Motor Assessment

AIM The aim of the study was to determine the best measure to discriminate between those with oropharyngeal dysphagia (OPD) and those without OPD, among young children with cerebral palsy (CP).

METHOD We carried out a cross-sectional population-based study involving 130 children with CP aged between 18 months and 36 months (mean 27.4mo; 81 males, 49 females) classified according to the Gross Motor Function Classification Scale (GMFCS) as level I (*n*=57), II (*n*=15), III (*n*=23), IV (*n*=12), or V (*n*=23). Forty children with CP (mean 28.5mo; 21 males,19 females, eight for each GMFCS level) were included in the reproducibility sub-study, and 40 children with typical development (mean 26.2mo; 18 males, 22 females) were included in the validity sub-study. OPD was assessed using the Dysphagia Disorders Survey (DDS), Pre-Speech Assessment Scale (PSAS), and Schedule for Oral Motor Assessment (SOMA). We analysed reproducibility using inter- and intrarater agreement (percentage) and reliability (kappa values and intraclass correlation coefficients). Construct validity was assessed as concordance between measures (SOMA, DDS, and PSAS). In the absence of a criterion standard measure for OPD, prevalence was estimated using latent class variable analysis. Data from the children with typical development were used to propose modified OPD cut-points for discriminative validity.

RESULTS All measures had strong agreement (>85%) for inter- and intrarater reliability. The SOMA had the best specificity (100.0%), but lacked sensitivity (53.0%), whereas the DDS and PSAS had high sensitivity (each 100.0%) but lacked specificity (47.1% and 70.6% respectively). OPD prevalence when calculated using the web-based estimation was 65.4%, which was similar to the estimate from the modified cut-points.

INTERPRETATION Using the sample of children with typical development and modified cut-points, OPD prevalence was lower than estimates with standard scoring. We propose using these modified cut-points when administering the DDS, PSAS or SOMA in young children with CP.

Oropharyngeal dysphagia (OPD) has been reported in 85% of children with cerebral palsy (CP)[1] and may result in limitations to dietary intake and nutritional status, as well as increasing children's risk of respiratory compromise.[2] CP describes a group of motor disorders resulting from a static brain lesion, which may impact on gross motor, fine motor, or oral sensorimotor control, leading to impairments in speech and feeding.[3] The term OPD is used in this study to describe impairment to any component of the oral-preparatory, oral (propulsive), and/or pharyngeal phases of the swallow, associated with eating, drinking, or controlling saliva.[4]

There is little consensus in the literature regarding the terms used to describe feeding impairments or mealtime activity limitations, and agreement on the construct parameters is lacking. The terms OPD, feeding/deglutition disorder, and oral motor dysfunction have been used frequently, but have variably included delayed and dysfunctional feeding; motor, sensory and/or behavioural feeding difficulties; oropharyngeal impairments and those to self-feeding (see Table SI, online supporting information, for a list of terms and definitions). This variability in the domains encompassed by these terms has in part led to the variability in results of studies of OPD. The construct of OPD is generally agreed to consist of three main phases, the oral, pharyngeal, and oesophageal phases,[5] with the oral phase sometimes further divided into the oral-preparatory and oral-propulsive phases.[4] The skills needed to ingest the

DOI: 10.1111/dmcn.12616

various food/fluid textures (broadly including spoonable foods, chewable foods, and fluids) are distinct but overlapping, including orienting to the approaching bolus, accepting the bolus (clearing a spoon, biting, or sipping from a cup or bottle), processing the bolus without anterior loss, propelling the bolus into the pharynx to initiate the swallow, and the swallow itself. Most OPD assessments cover at least these three broad textures, and ability to process these textures therefore represent a minimum skill set for feeding evaluation (see clinimetric review for details of OPD assessments).[6]

In order to optimally manage OPD in children with CP, measures with strong validity, reproducibility, and clinical utility are needed to allow accurate identification of OPD case status and severity. OPD can be measured using instrumental methods such as videofluoroscopy, but initial assessment usually uses direct objective measures in the clinic. After a systematic review,[6] the Schedule for Oral Motor Assessment (SOMA)[7] and Dysphagia Disorders Survey (DDS)[8] were identified as two measures with the strongest psychometric properties for use in young children with neurodevelopmental disabilities. The Pre-Speech Assessment Scale (PSAS)[9] was considered a comprehensive measure that is used widely in the clinical setting. These three measures were selected for use in a prospective cohort study of children with CP aged 18 to 36 months[10] and were therefore the subject of the current analysis.

In previous work, 40% of children were identified as having OPD based on the SOMA, whereas this figure was almost doubled (78%) when these same children were assessed using the DDS.[1] The SOMA has been developed through seeded cluster analysis of children with typical development, children with non-organic failure to thrive, and a small group of children with CP and clinically significant feeding difficulties (n=13). This methodology for development of the measure means that the SOMA is likely to have a high degree of specificity (i.e. children identified as having OPD on the SOMA are likely to have 'true' OPD); however, the sensitivity, particularly for mild OPD, has not been adequately explored. The DDS was developed primarily in adults with developmental disability and, although it has been validated for children as young as 2 years, there has been inadequate sampling of children of this age.[11] Thus, it is possible the DDS is overdetecting OPD in children with newly established oral sensorimotor systems. Arising from this literature review[6] and preliminary study,[1] this study aims to determine the best measure to accurately discriminate those with OPD from those without OPD among young children with CP. It is hypothesized that the SOMA will accurately identify children with OPD but will miss cases with mild OPD, and the DDS would overdetect OPD cases associated with typical development.

METHOD

This is a cross-sectional analysis of a population-based study of preschool children with CP and same-aged children with typical development. It forms part of two concurrent longitudinal studies of motor function and brain structure (National Health and Medical Research Council 465128) and growth, nutrition, and physical activity (National Health and Medical Research Council 569605).[10,12,13] Ethics approval was provided by the University of Queensland Medical Research Ethics Committee (2008002260), the Children's Health Services District Ethics Committee (HREC/08/QRCH/112), and at other regional and organizational ethics committees, with an amendment to include a reference sample (children with typical development). All families gave written informed consent to participate.

What this paper adds

- The Dysphagia Disorders Survey (DDS) and Pre-Speech Assessment Scale (PSAS) had high sensitivity in detecting oropharyngeal dysphagia (OPD), but low specificity. In contrast, the Schedule for Oral Motor Assessment (SOMA) had high specificity, but low sensitivity.
- OPD prevalence is approximately 65% when using web-based estimation methods, and with modified OPD cut-points.
- Modified cut-points should be used for classifying OPD on the DDS, PSAS, and SOMA in young children with CP. This will improve the specificity of the measures.
- DDS, PSAS, and SOMA had strong inter- and intrarater reproducibility.

Participants

Three samples were recruited for this study: the growth, nutrition, and physical activity study sample; the reproducibility sample; and a sample of children with typical development (Fig. S1, online supporting information). Children with a confirmed diagnosis of CP, aged 18 to 36 months (corrected age) at the time of initial assessment, and born in Queensland between 2006 and 2009, were invited to participate in the study.[12] Children with neurodegenerative conditions were excluded. Ten additional children with CP age 18 to 36 months (corrected age), from all birth places and birth years, were recruited to participate in the reproducibility sample (resulting in four children from each Gross Motor Function Classification System [GMFCS] level in each of the 18–24mo and 30–36mo age bands). These additional children were recruited for pragmatic reasons related to another substudy associated with this work.

Forty children with typical development were recruited through convenience sampling for the discriminative validity substudy, stratified by age (20 children aged 18–24mo, 20 children aged 30–36mo). The children were term born (at ≥37wks), did not have a diagnosis which required neonatal admission or ongoing medical or allied health treatment, and were not on regular medication.

Measures

Three clinical measures of OPD were evaluated in this study (SOMA, DDS, and PSAS). Their psychometric properties have been detailed in previous work.[6,10] The SOMA is a discriminative measure identifying oral motor dysfunction in children according to skills typically mastered from 8 to 24 months.[7] It categorizes oral motor dysfunction into

a binary yes/no outcome using cutting scores defined for each of seven oral motor challenge categories (purée, semi-solid, solid, cracker, bottle, trainer cup, cup).[7] The SOMA has been validated on 127 young children: 58 comparison children with typical oral skills, 56 with non-organic failure to thrive (aged 8–24mo), and 13 children with CP and overt feeding difficulties (aged up to 42mo).[14]

The DDS-Pediatric (Part 2) is an evaluative measure for screening signs of oral, pharyngeal, and oesophageal phase dysphagia in children and adults with developmental disability.[8] Part 2 provides a raw score based on a series of binary judgements that indicates an individual's functional eating competency (maximum impairment raw score of 22).[15] The DDS underwent final standardization on 427 individuals with developmental disability (mean age 33y),[15] with the paediatric measure validated in a group of 166 children (range 2y 1mo–19y 1mo; mean 9y 4mo) with moderate to severe CP (GMFCS III–V) and intellectual disability.[15]

The PSAS is an evaluative measure that examines 27 pre-speech feeding behaviours up to 24+ months, according to performance areas of sucking, swallowing, biting, chewing, respiration-phonation, and sound play.[9] Only subtests pertaining to the ingestion of food/fluid were administered (sucking, swallowing, biting/chewing). Each item is scored on an ordinal 'disorder' scale (maximum of nine), and a developmental scale (with age norms, up to 24+ months), to provide a double score overall. The PSAS was developed through a 3-year longitudinal study of six children, and field testing of the measure for 8 years by 215 trained clinicians, who provided annual feedback on its clinical use.[9]

Children's gross motor function was classified as one of five levels using the GMFCS by two physiotherapists. The age bands '<2 years' and '2-4 years' were used.[16]

Statistical analysis

Three main analyses were completed to assess the psychometric properties of the measures: reproducibility, construct validity, and discriminative validity. For reproducibility, inter- and intrarater agreement was calculated using percentage agreement. Binary measures were defined to agree if they had perfect agreement, and measures with more than five categories/points considered to agree if they were within 1 unit. For these multi-categorical variables, the 95% limits of agreement were also calculated. Reliability was assessed using kappa statistics and intraclass correlation coefficients. For construct validity, the concordance between the three measures overall, and pairwise concordance between the SOMA, DDS, and PSAS, were calculated. In the absence of a criterion standard measure, the overall prevalence, sensitivity, and specificity of each measure were estimated using latent class variable analysis, whereby initial estimates of prevalence, sensitivity, and specificity were refined using maximum likelihood methods.[17] Calculations were conducted using the TAGS (Tests in the Absence of Gold Standard) internet-based software (UC Davis, Davis, CA, USA).[17] For discriminative validity, data from the children with typical development were presented descriptively for each measure, and the specificity of measures calculated based on the assumption that children with typical development are all true negative cases.

Modified cut-points for the classification of OPD in young children with CP were calculated based on the mean score of the children with typical development plus two standard deviations (–2SD for the PSAS). This identified the score threshold which will be exceeded by only 2.5% of children with typical development (for each age stratum, 18–24mo and 30–36mo). Modified cut-points were used to calculate the proportion of children with CP with significantly higher scores than children with typical development. The discriminative validity of each measure's items was calculated using binomial logistic regression, with sample (typical development/CP) as the main effect. Statistical analyses were performed using Stata v10.0 (StataCorp, College Station, TX, USA).

RESULTS

A total of 130 children participated in the growth, nutrition, and physical activity study (81 males, 49 females; mean age 27.4mo). Forty children with typical development (18 males, 22 females; mean age 26.2mo) and 10 additional children with CP were recruited only to the reproducibility study (giving a total reproducibility of 40 stratified for age [18–24mo, *n*=20; and 30–36mo, *n*=20] and GMFCS [*n*=8 per GMFCS level], 21 males, 19 females; mean age 28.5mo). The recruitment pathways of the three samples can be found in Figure S1. The full sample characteristics are presented in Table I.

Table I: Participant characteristics

	GNPA (*n*=130)	TD (*n*=40)
Age (mo), mean (SD)	27.4 (5.4)	27.2 (5.9)
Sex, males, *n* (%)	81 (62.3)	18 (45.0)
GMFCS level, *n* (%)		NA
I	57 (44.2)	–
II	15 (11.6)	–
III	23 (17.8)	–
IV	12 (9.3)	–
V	23 (17.7)	–
Primary motor type, *n* (%)		NA
Unilateral spasticity	41 (31.5)	–
Bilateral spasticity	72 (55.4)	–
Dystonia	2 (1.5)	–
Ataxia	2 (1.5)	–
Hypotonia	9 (6.9)	–
Athetoid	4 (3.1)	–
Motor distribution, *n* (%)		NA
One limb	2 (1.6)	–
Two limbs	67 (51.5)	–
Three limbs	13 (10.0)	–
Four limbs	48 (36.9)	–
Tube fed, *n* (%)		
Partial	11 (8.4)	0.0 (0.0)
Complete	5 (3.9)	0.0 (0.0)

GFCS, Gross Motor Function Classification System; GNPA, growth, nutrition, and physical activity; NA, not applicable; SD, standard deviation.

Oropharyngeal dysphagia in children with cerebral palsy and those with typical development

The proportion of children (those with CP and those with typical development) with a positive OPD case classification on the DDS, SOMA, and PSAS and their corresponding scores are shown in Table II. Children with CP, as a group, consistently had a greater proportion and severity of OPD than children with typical development ($p<0.01$).

Reproducibility of oropharyngeal dysphagia measures

The reliability and agreement for inter- and intrarater were significant for all variables (Table III). The interrater agreement was >85% for all binary measures and intrarater agreement >90%. The mean of differences between raters for the DDS was 0 (SD 2.6), for PSAS delay was –1.9 (SD 2.5), and for PSAS disorder was 0.3 (SD 1.0). The intrarater mean of differences for the DDS was 0.3 (SD 1.4), for PSAS delay was –0.2 (SD 2.7), and for PSAS disorder was 0.1 (SD 0.7).

Convergent validity of oropharyngeal dysphagia measures

The three measures (SOMA, DDS, and PSAS) were triangulated, with their agreement shown in Figure 1. The agreement of measures for the CP sample and sample of children with typical development was used for the prevalence calculation using latent class variable analysis. A 'best guess' prevalence estimate of 80% was used, based on previous research by our team.[1] Sensitivity and specificity estimates for each measure were based on previous test use and the results from the sample of children with typical development (DDS: sensitivity=0.99, specificity=0.50; SOMA: sensitivity=0.50, specificity=1.00; PSAS: sensitivity=0.99, specificity=0.63). The OPD prevalence estimate obtained from this estimation was 65.4%, with sensitivity and specificity estimated as follows: SOMA, sensitivity=53.0%, specificity=100.0%; DDS, sensitivity=100.0%, specificity=47.1%; and PSAS, sensitivity=100.0%, specificity=70.6%.[17]

Discriminative validity of oropharyngeal dysphagia measures

Using the modified cut-points based on the scores of children with typical development (mean ±2SD), the proportion of children with OPD was 56.6% for the DDS, 62.2% for the SOMA, and 45.5% for the PSAS (Fig. 2).

Table II: Proportion of children with positive oropharyngeal dysphagia classification and scores on OPD measures for children with typical development and children with cerebral palsy

	Dysphagia Disorders Survey (DDS)			
	% with OPD	Mean (SD)	Range	Specificity
TD	50.0 (42.0–65.8)	0.8 (1.1)	0–4	50.0
CP overall	84.6 (83.3–90.9)[a]	7.1 (7.3)[a]	0–22	
GMFCS I	66.7 (54.2–79.1)	2.4 (2.9)[a]	0–11	
GMFCS II	100.0 (78.2–100.0)[a]	3.9 (3.8)[a]	1–15	
GMFCS III	95.7 (70.7–100.0)[a]	6.8 (4.4)[a]	0–15	
GMFCS IV	100.0 (73. 5–100.0)[a]	12.1 (7.8)[a]	1–22	
GMFCS V	100.0 (85.2–100.0)[a]	19.1 (2.7)[a]	14–22	
	Schedule for Oral Motor Assessment (SOMA)			
	% with OPD	Mean (SD)[b]	Range	Specificity
TD	0.0 (0.0–8.8)	0.7 (1.0)	0–5	100.0
CP overall	34.6 (26.3–42.9)[a]	4.7 (7.4)[a]	0–29	
GMFCS I	8.8 (1.3–16.2)[a]	1.9 (3.0)	0–12	
GMFCS II	13.3 (4.6–31.3)[a]	2.6 (3.4)	0–12	
GMFCS III	26.1 (7.6–44.6)[a]	8.2 (7.8)[a]	1–21	
GMFCS IV	75.0 (49.2–100.0)[a]	NA	NA	
GMFCS V	100.0 (85.2–100.0)[a]	NA	NA	

	Pre-Speech Assessment Scale (PSAS)					
	% with OPD	Mean delay (SD)[c]	Mean dysfunction score (SD)	Delay (range)	Dysfunction score (range)	Specificity
TD	37.5 (22.2–52.8)	0.1 (2.1)	0.0 (0.0)	–4.2 to 6.2	0–0	62.5
CP overall	72.9 (65.1–80.6)[a]	7.1 (8.5)[a]	1.0 (2.8)[a]	–2.2 to 25.0	0–9	
GMFCS I	55.4 (42.1–68.6)	2.0 (3.4)[a]	0.1 (0.5)	–2.2 to 15.5	0–3	
GMFCS II	66.7 (41.8–91.5)	3.0 (4.3)[a]	0.4 (1.1)	–1.9 to 12.3	0–4	
GMFCS III	78.3 (60.9–95.6)[a]	6.5 (4.9)[a]	0.3 (0.6)	0.4 to 14.6	0–2	
GMFCS IV	100.0 (73.5–100.0)[a]	13.0 (8.7)[a]	3.7 (3.1)[a]	1.0 to 21.0	0–8	
GMFCS V	100.0 (85.2–100.0)[a]	21.4 (4.0)[a]	6.6 (1.9)[a]	10.7 to 25.0	0–9	

Specificity of measures based on the assumption that children with typical development have no OPD. [a]Significant difference in GMFCS level compared with children with typical development sample based on logistic regression (proportion) and linear regression (score). [b]Mean SOMA score based on sum of purée, chewable, and cup raw scores (n=85 with scores in these fields). NA indicates four or fewer observations at this GMFCS level. [c]Delay is equal to age in months minus 'delay' score. CP, cerebral palsy; OPD, oropharyngeal dysphagia; GMFCS, Gross Motor Function Classification System; SD, standard deviation; TD, typical development.

Table III: Reproducibility (interrater and intrarater) of oropharyngeal dysphagia measures in preschool children with cerebral palsy

	Interrater (*n*=40)		Intrarater (*n*=40)	
	Reliability (κ)	% Agreement	Reliability (κ)	% Agreement
SOMA (overall)	0.7[a]	85.0	0.9[a]	92.5
Purée	0.7[a]	86.1	0.9[a]	94.4
Semi-solid	0.9[a]	96.7	1.0[a]	100.0
Solid	1.0[a]	100.0	1.0[a]	100.0
Cracker	0.8[a]	91.2	0.8[a]	90.9
Bottle	0.8[a]	90.0	1.0[a]	100.0
Trainer cup	0.9[a]	95.8	1.0[a]	100.0
Cup	0.9[a]	96.3	1.0[a]	100.0
DDS-Part 2 (overall)	0.7[a]	97.5	0.4[a]	92.5
DDS-Part 2 (raw score)[b]	0.92[a]	39.5	0.96[a]	75.0
Non-chewable score[b]	0.95[a]	94.9	0.95[a]	90.0
Chewable score[b]	0.70[a]	69.2	0.95[a]	95.0
Fluid score[b]	0.75[a]	84.6	0.92[a]	92.5
PSAS (overall)	0.5[a]	95.0	0.5[a]	92.5
PSAS delay (binary)	0.8[a]	97.5	0.5[a]	92.5
PSAS delay (score)[b]	0.99[a]	38.2	0.98[a]	70.0
PSAS disorder (binary)	0.5[a]	76.9	0.8[a]	87.5
PSAS disorder (score)[b]	0.93[a]	88.2	0.99[a]	95.0

[a]*p*-value <0.01; κ<0, poor; 0.01–0.20, slight; 0.21–0.40, fair; 0.41–0.60, moderate; 0.61–0.80, substantial; and 0.81–1.0, almost perfect.[19]
[b]Multi-categorical variables, therefore intraclass correlation coefficient reported and per cent agreement is perfect agreement and 1 point either side. DDS, Dysphagia Disorders Survey; κ, Cohen's kappa coefficient; OPD, oropharyngeal dysphagia; PSAS Pre-Speech Assessment Scale; SOMA, Schedule for Oral Motor Assessment.

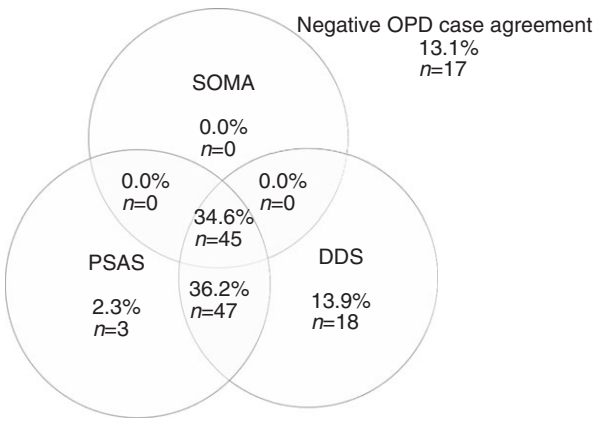

Figure 1: Agreement between measures of oropharyngeal dysphagia (OPD) in preschool children with cerebral palsy. DDS, Dysphagia Disorders Survey; PSAS, Pre-Speech Assessment Scale; SOMA Schedule for Oral Motor Assessment.

The items with poor discrimination between the sample of children with typical development and mild CP (GMFCS I–II) are presented in Table SII, online supporting information. A large number of children with typical development scored on the post-swallow items of the DDS (suggesting these items may be overdetecting typical feeding patterns in young children). Further, reception, containment, and oropharyngeal swallow (lack of sequential swallows) for fluids on the DDS were also detected in the children with typical development. The trainer cup and cup subtests of the SOMA discriminated poorly between the children with typical development and those with mild CP owing to potentially poor detection of mild OPD on these subtests, as was the case for the swallowing subtests on the PSAS.

DISCUSSION

The overarching aim of this study was to determine which measure could most accurately estimate OPD prevalence in preschool children with CP. This arose out of variability in the estimates of the measures when used in previous work by our team.[1] The prevalence of OPD in young children with CP may be between 45% and 65% based on our representative population-based cohort (45% representing the lowest modified prevalence estimate, yielded by the PSAS, and 65% generated from the web-based estimation). The inclusion of a third measure (the PSAS) and use of the children with typical development to provide further validation of the DDS, SOMA, and PSAS has provided valuable information in understanding the performance of the measures in young children with CP.

Based on previous work by our team[1] and the current study, the reproducibility of the SOMA, DDS, and PSAS (as binary measures) is considered strong. The use of scores on the DDS and PSAS to indicate OPD severity should be carefully interpreted, depending on each specific use (e.g. to describe change in a child when used by a consistent rater, or to compare children between settings with different raters). The accuracy of the DDS was high, within approximately 2 points between repeated measurements with a consistent rater, but was accurate to only 4 points either side between two certified raters. The PSAS scores performed similarly whether with a consistent rater or between raters, accurate to about 5 months either side of the age estimate, which would also be a clinically meaningful threshold. Previous published work on the reproducibility

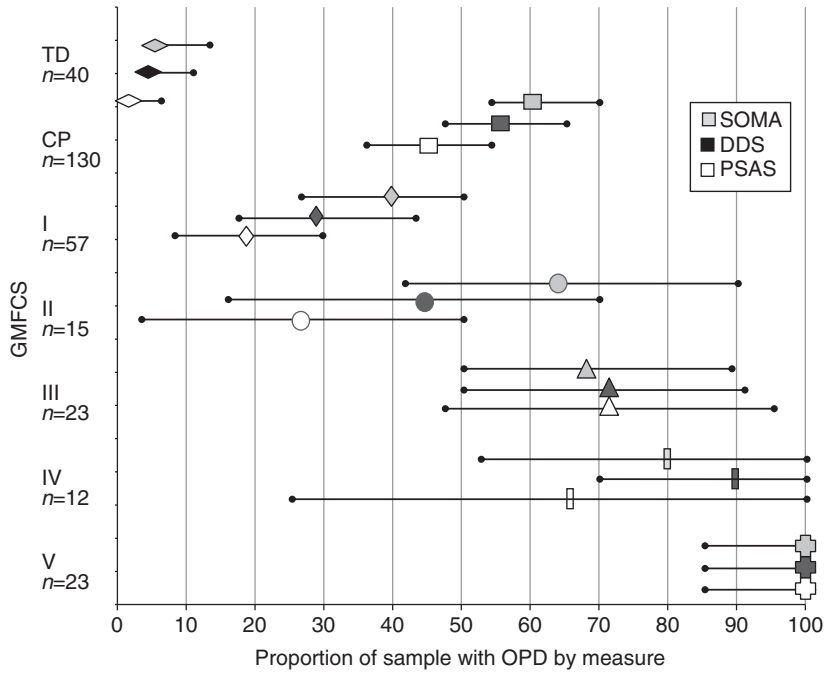

Figure 2: Modified cut-points of oropharyngeal dysphagia (OPD) based on typically developing reference data and recalculated prevalence estimates for young children with cerebral palsy. Mean calculated for each subtest (by age), with oral motor dysfunction rating on one or more subtests constituting OPD overall. SOMA modified cut-points are as follows: purée, 18–24mo, 0; 30–36mo, ≥1; semi-solid, 18–24mo, 0; 30–36mo, 0; cracker, 18–24mo, ≥1; 30–36mo, ≥3; trainer cup, 18–24mo, ≥2; 30–36mo, 0; cup, 18–24mo, ≥2; 30–36mo, ≥2. DDS modified cut-points are ≥4 for 18–24mo and ≥3 for 30–36mo; PSAS modified cut-point is delay score ≤4mo compared with age. CP, cerebral palsy; DDS, Dysphagia Disorders Survey; GMFCS, Gross Motor Function Classification System; PSAS, Pre-Speech Assessment Scale; SOMA, Schedule for Oral Motor Assessment.

of these measures was also reported to be strong (97% agreement for the DDS, 65–87% for the PSAS, and perfect agreement on 56–68% of items on the SOMA), although the number of studies and methodological rigor were limited.[6] Except in our previous work,[1] intrarater reliability was not tested for any of the measures, and only a single study reported on interrater reliability for the DDS and PSAS, and two small studies for the SOMA.

In the present study, we recruited a sample of children with typical development to obtain information regarding measure performance in an assumed 'OPD-free' sample. Strikingly, half the children with typical development were identified as having OPD on the DDS, and 37% on the PSAS, suggesting that the specificity of these measures is relatively poor (i.e. they are identifying children as having OPD for children who may be within the range of normal). While these figures suggest an overdetection when using a binary 'OPD/not' judgement, when difference in scores are analysed, we see consistently higher scores for the CP sample, even between children in GMFCS level I and those with typical development. This supports the use of the DDS raw score to indicate OPD severity, but suggests that existing cut-points for the classification of OPD may be too sensitive. Children with typical development demonstrated only a small number of items on the DDS: post-swallow on all textures (i.e. cough or wet breath), difficulty sipping from a cup, dribbling fluids when drinking from a cup, and not demonstrating sequential fluid swallows. Impairment on these same items was frequently seen in children with ambulatory CP, but in a greater proportion of children classified as GMFCS levels II and III (see Table SII, online supporting information).

Based on the combined results of the TAGS software and using modified cut-points, the prevalence estimate for this representative sample of young children with CP was approximately 60% (although could plausibly be as low as 45% and up to 65%). Further, based on these combined results, we can say with a fair degree of confidence that all children classified as GMFCS level V will have OPD. There was consensus from all three measures that feeding performance in children with CP is significantly poorer than in children with typical development, even among children whose motor function is classified as GMFCS level I. Using the modified cut-points, our prevalence estimate in the GMFCS levels IV–V group has changed minimally, to around 90%, which is still consistent with the prevalence estimates presented in the literature previously for children of these GMFCS levels.[15,18] Through the inclusion of the sample of children with typical development, we propose that OPD detected on the DDS and PSAS has included some background noise of 'limitations' to ingestion functions that are part of typical development.

Selecting the most appropriate measure for detecting OPD in young children with CP is critical for future research and optimal service planning. On all three measures, there was strong agreement among raters for detecting OPD, and so one measure should not be prioritized for use based on this property. The PSAS and DDS were in agreement regarding case status about 85% of the time, which suggests they are both capturing a similar construct of OPD. Both the present study and previous literature have suggested the SOMA is detecting clinically significant OPD, but lacks sensitivity to detect milder cases. It could be argued that this clinically significant OPD is the construct with which we should be concerned, with regards to potential impact on health outcomes. However, the association between various constructs of OPD and health outcomes have not been well demonstrated in the literature (particularly as assessed by standardized measures), and we therefore advocate use of a measure with greater sensitivity (i.e. the DDS or PSAS) to ensure that we are detecting the true extent of the impairment. Further work is needed to determine whether the construct these measures are capturing is the one that is most clinically relevant (based on impact on health and treatability). When using one of these more sensitive measures, we do not want to grossly overdetect OPD by including 'limitations' to ingestion functions associated with typical development. For this reason we recommend using our modified cut-points when applying these measures to young children with CP, to improve their specificity. Despite the fact that the PSAS was found to have better specificity than the DDS when calculated using the TAGS software, we would support the use of the DDS with modified cut-points as the best measure available for determining OPD in young children with CP. The scoreable version of the PSAS is no longer in use, and, although the PSAS checklists are useful in the clinical setting, there are no cut-points to guide practice decision, and this is less useful for research purposes. In the case of the DDS, the measure is still published, the clinical decisions required for scoring are more specific (which would mean that its reliability would likely remain strong even when used by less familiar users), its scoring structure is more systematic and easily interpreted (i.e. each texture covers the same eight ingestion functions, except for exclusion of the 'chewing' item for non-chewable foods and fluids), and an increase in score reflects a linear increase in the extent of the functional impairment.

The children who were detected as having OPD on the SOMA (n=45) are almost certainly true cases of OPD, and those detected as not OPD on the DDS (n=20) are almost certainly true non-cases of OPD. This leaves us with 50% of the sample whose OPD classification is less definitive, some of whom may have mild OPD, some who may have delayed oropharyngeal sensorimotor skills, and some who may be false positives. Using the SOMA with its current scoring, the children we need to be most concerned about from a safety perspective may be detected, but it may be missing children who could still benefit from direct sensorimotor treatments (e.g. to work on the speed, strength, and coordination of specific movements such as biting, tongue lateralization, or cup drinking). The DDS and PSAS estimates are likely to include not only 'disordered' feeding, but also 'delayed' feeding, which reiterates the question of what the construct of OPD is deemed to encompass. Our future work analysing the longitudinal changes to the prevalence of OPD in our sample of young children with CP will assist in understanding the potential OPD subgroups.

This study has presented novel data on the psychometrics of the DDS, PSAS, and SOMA when used in young children with CP. While our sample of 130 children was substantial, when analysing data by GMFCS level, we had broad confidence intervals because of the small numbers and the between-child heterogeneity in the mid-range GMFCS levels (II–IV). The largest limitation in this study, however, was the lack of a criterion standard measure. Perhaps with greater time and investment, an expert speech pathology panel could serve as the criterion standard. This would particularly support our understanding of the sensitivity of the measures, which in the present study could be estimated only using the statistical models of the TAGS software. It is expected, however, that this study has provided greater clarity surrounding the construct of OPD in children with CP, and that our future work will further contribute to delineating this varied diagnosis.

ACKNOWLEDGEMENTS

This project was supported by the National Health and Medical Research Council Postgraduate Medical and Dental Scholarship (1018264 – KAB), Career Development Fellowship (APP1037220 – RNB) and Project Grants (569605 and 465128). Funding was also received from the Speech Pathology Australia Post Graduate Student Research Grant to conduct reliability ratings. We would like to thank physiotherapists Rachel Jordan (BPT) and Chris Finn (BPT) for data collection and gross motor ratings; and Dieticians Stina Oftedal (BHlthSc [Hons] Nutr & Diet) and Camilla Davenport (BHlthSc [Hons] Nutr & Diet) for data collection of feeding videos. The authors have stated that they had no interests that could be perceived as posing a conflict or bias.

SUPPORTING INFORMATION

The following additional material may be found online:

Figure S1: Recruitment samples for reproducibility and validity of measures of oropharyngeal dysphagia study

Table SI: Terms and construct of oropharyngeal dysphagia

Table SII: Dysphagia disorders survey sub-tests and items with poor discriminative validity between typically developing children and GMFCS I/II

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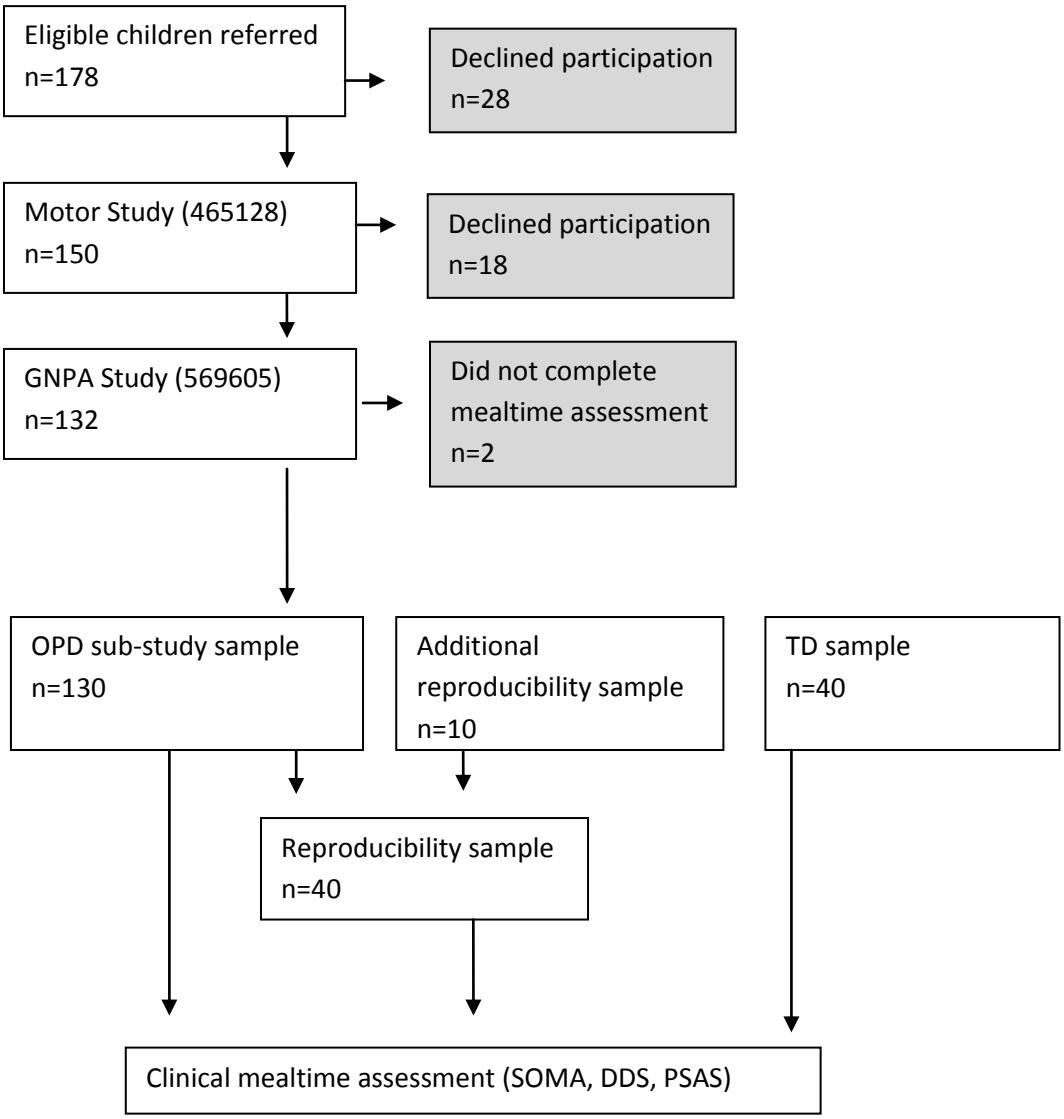
Table SI: Terms and construct of oropharyngeal dysphagia

Term/ Model	Definition
Eating[1]	*Carrying out the coordinated tasks and actions of eating food that has been served, bringing it to the mouth and consuming it in culturally acceptable ways, cutting or breaking food into pieces, opening bottles and cans, using eating implements, having meals, feasting or dining.*
Drinking[1]	*Taking hold of a drink, bringing it to the mouth and consuming the drink in culturally acceptable ways, mixing, stirring and pouring liquids for drinking, opening bottles and cans, drinking through a straw or drinking running water such as from a tap or a spring, feeding from the breast.*
Ingestion functions[1]	*Functions related to taking in and manipulating solids or liquids through the mouth into the body.*
Swallowing[1]	*Functions of clearing the food and drink through the oral cavity, pharynx and oesophagus into the stomach at an appropriate rate and speech. Includes oral, pharyngeal and oesophageal swallowing.*
Oropharyngeal Dysphagia[2]	*Characterised by problems in any or all phases of swallowing (bolus formation, oral transit, pharyngeal phase, upper-oesophageal phase).*
Dysphagia[3]	*A swallowing disorder. The signs and symptoms of dysphagia may involve the mouth, pharynx, larynx, and/or esophagus.*
Dysphagia[4]	*Characterised by deficiencies in oral preparation, oral-pharyngeal and oesophageal phases of swallowing. It can be caused by oromotor dysfunction, anatomical anomalies, abnormal neurological maturation, oral sensory impairment or oesophageal motility disorders.*
Feeding disorder[3]	*Disordered placement of food in the mouth; difficulty in food manipulation prior to initiation of the swallow, including mastication; and the oral stage of the swallow when the bolus is propelled backward by the tongue. In pediatrics, this term may be used to describe a failure to develop or demonstrate developmentally appropriate eating and drinking behaviors.*
Feeding disorder[5]	*Problems in a broad range of eating activities that may or may not be accompanied by a difficulty with swallowing food and liquid. Feeding disorders may be characterised by food refusal, disruptive mealtime behaviour, rigid food preferences, less than optimal growth, and failure to master self-feeding skills expected for developmental levels.*
Swallowing and feeding disorders[6]	*Dysphagia and delays and/ or disorders in the development of eating and drinking skills. Includes the introduction, preparation, transfer, and transport of food and liquid from mouth through oesophagus into stomach; also includes management of saliva and oral intake of medications.*
Deglutition disorders[7]	MeSH term: *Difficulty in swallowing which may result from neuromuscular disorder or mechanical obstruction. Dysphagia is classified into two distinct types: oropharyngeal dysphagia due to malfunction of the pharynx and upper oesophageal sphincter; and esophageal dysphagia due to malfunction of the oesophagus.*
Process Model of Feeding/ Swallow Phases[8]	Oral preparatory: After food/ liquid enters mouth, the ability to hold the bolus through closure of the lips, soft palate and tongue contact. Oral propulsive: Movement of the bolus from anterior to posterior oral cavity. Includes 'food processing' stage for solid boluses. Pharyngeal: Bolus passage through the pharynx and upper oesophageal sphincter to the oesophagus. Includes airway closure/ protection.

	Oesophageal: Bolus passage to the stomach through peristaltic muscle contraction.
Swallowing disorders[5]	As for 'oropharyngeal dysphagia' (above).
Medical, oral (motor, sensory), behavioural[9]	*Medical: specific medical diagnosis.* *Oral: any oropharyngeal functional abnormality (including sensory or motor).* *Behavioural: when current norms and rules were crossed in a specific situation.*
Structural, neurodevelopmental, behavioural[10]	(1) Structural abnormalities (nasopharynx, larynx, trachea, oesophagus) (2) Neurodevelopmental feeding disorders: *related to 'learning to eat' resulting in oral hypersensitivity and oral-motor dysfunction.* (3) Behavioural feeding disorders (DSM-IV-TR).
Structural, neurological, behavioural[11]	*Structural: abnormalities to structures associated with eating & drinking* *Neurological: feeding problems associated with central nervous system/ musculoskeletal disorders.* *Behavioural: resulting from psychosocial difficulties, negative feeding behaviours or emotionally-based difficulties.* *Cardiorespiratory: feeding difficulties associated with diseases of the cardiovascular/ respiratory systems.* *Metabolic: feeding difficulties associated with metabolic conditions.*
Eating impairment[12]	*Combination of growth and eating skills.*
Eating skills[12]	*Includes eating efficiency and oral motor skills.*
Eating efficiency[12]	*Child's ability to ingest a nutritionally adequate diet and consume enough calories within a reasonable amount of time to permit growth within normal limits* (standard age-based curves exist).
Eating competence/ oral motor skills[12]	*Traditionally includes jaw and lip control, tongue mobility , chewing vigor, drinking skills, and safety of swallowing.*
Oral-Motor Dysfunction[13]	*Concomitant of certain congenital anomalies (e.g. cleft palate) or neurological disorders (e.g. bulbar palsy), and delay in the development of appropriate skills is associated with mental retardation.*

Italicised for directly quoted definitions; [1]World Health Organization. International Classification of Functioning, Disability and Health: Online version. 2011 [cited 2011 26 September]; Available from: http://apps.who.int/classifications/icfbrowser/Default.aspx; [2]Arvedson JC. Feeding children with cerebral palsy and swallowing difficulties. Eur J Clin Nutr [serial on the Internet]. 2013; 67(S9-S12): Available from: http://www.nature.com/ejcn/journal/v67/n2s/full/ejcn2013224a.html; [3]American Speech-Language-Hearing Association. Roles of speech-language pathologists in swallowing and feeding disorders: Technical report2001: Available from: www.asha.org/policy; [4]Calis EA, Veugelers R, Sheppard JJ, Tibboel D, Evenhuis HM, Penning C. Dysphagia in children with severe generalized cerebral palsy and intellectual disability. Dev Med Child Neurol. 2008;50:625-30; [5]Arvedson JC. Assessment of pediatric dysphagia and feeding disorders: clinical and instrumental approaches. Developmental Disabilities Research Reviews. [Review]. 2008;14:118-27; [6]American Speech-Language-Hearing Association. Guidelines for speech-language pathologists providing swallowing and feeding services in schools2007: Available from: www.asha.org/policy; [7]Medical subject headings: MeSH descriptor data [database on the Internet]2014. Available from: http://www.nlm.nih.gov/cgi/mesh/2014/MB_cgi; [8]Matsuo K, Palmer JB. Anatomy and physiology of feeding and swallowing -- normal and abnormal. Phys Med Rehabil Clin N Am. 2008;19:691-707; [9]Rommel N, De Meyer A-M, Feenstra L, Veereman-Wauters G. The Complexity of Feeding Problems in 700 Infants and Young Children Presenting to a Tertiary Care Institution. J Pediatr Gastroenterol Nutr. 2003;37:75-84; [10]Bernard-Bonnin A. Feeding problems of infants and toddlers. Can Fam Physician. 2006;52:1247-51; [11]Burklow KA, Phelps AN, Schultz JR, McConnell K, Rudolph C. Classifying Complex Pediatric Feeding Disorders. J Pediatr Gastroenterol Nutr. 1998;27:143-7; [12]Gisel E, Alphonce E. Classification of eating impairments based on eating efficiency in children with cerebral palsy. Dysphagia. 1995;10:268-74; [13]Mathisen B, Skuse D, Wolk D, Reilly S. Oral-motor dysfunction and failure to thrive among inner-city infants. Dev Med Child Neurol. 1989; 31:293-302

Figure S1: Recruitment samples for Reproducibility and Validity of Measures of Oropharyngeal Dysphagia Study



Key: DDS Dysphagia Disorders Survey; GNPA Growth, Nutrition and Physical Activity (study); OPD oropharyngeal dysphagia; PSAS Pre Speech Assessment Scale; SOMA Schedule for Oral Motor Assessment; TD Typically Developing

Table SII: Dysphagia disorders survey sub-tests and items with poor discriminative validity between typically developing children and GMFCS I/II

	TD	CP: GMFCS				
		I	II	III	IV	V
DDS						
Non-chewable (mean±SD)[†]	0.0±0.2	0.4±0.8	0.5±1.3	1.2±1.5*	3.2±2.7*	6.1±0.9*
Orientation (%±CI)[†]	0.0	0.0	6.7±10*	0.0	36.4± 10*	95.6±10 *
Post-swallow (%±CI)	2.5±5	10.9±8.4	0.0	13.6±14.8	36.4±30.4*	62.5±25.0*
Chewable post-swallow (%±CI)[α]	7.5±8.4	14.3±9.4	20.0±21.4	21.7±17.4	36.4±30.4*	62.5±25.0*
Fluids (mean±SD)[†]	0.6±0.9	0.9±1.2	1.4±1.4	2.1±1.9*	3.8±2.5*	6.0±1.0*
Orientation (%±CI)[†]	0.0	0.0	6.7±13.2*	4.8±9.4*	36.4±30.4*	73.3±23.6*
Reception (%±CI)[α]	5.0±6.9	14.5±9.4	26.7±23.6*	38.1±21.6*	72.7±28.0*	93.3±13.3*
Containment (%±CI)[α]	17.5±12.0	14.5±9.4	40.0±26.2*	14.3±15.6	63.6±30.4*	86.7±18.2*
Oral-pharyngeal (%±CI)[α]	5.0±7.0	12.7±9.0	13.3±18.1	28.6±20.2*	72.7±28.2*	93.3±13.3*
Post-swallow (%±CI)[α]	37.5±15.5	32.7±12.8	40.0±26.2	28.6±20.2	36.4±30.4	60.0±26.2
SOMA						
Trainer cup (%±CI)[†]	0.0	0.0	12.5±25.0*	0.0	66.7±66.7*	100.0±0.0*
Cup (%±CI)[†]	0.0	0.0	0.0	0.0	0.0	100.0±0.0*
PSAS						
Sucking (mean, SD)[†]	0.3±2.8	1.9±4.2	3.3±4.1*	4.8±3.2*	9.9±7.6*	19.7±5.4*
Bottle[†]	-1.7±2.3	-0.1±4.2	-1.7±2.7	-0.8±3.2	6.1±9.1*	13.8±12.8*
Cup[†]	-0.7±2.8	1.0±4.0	2.1±6.5*	5.3±5.4*	13.9±9.7*	21.7±3.9*
Swallowing (mean, SD)[†]	0.2±2.4	1.9±4.4	2.0±4.6	7.5±6.2*	13.0±7.7*	21.8±3.5*
Solids[†]	-1.0±3.1	1.5±5.1*	0.5±4.2	6.4±9.4*	9.1±9.8*	20.8±5.1*
Coordination[α]	2.5±6.0	2.9±6.6	2.3±6.1	9.8±10.2*	13.3±10.5*	21.6±4.4*
Bite chew	NA	NA	NA	NA	NA	NA
Jaw bite	-0.1±3.4	2.3±5.0*	3.0±6.4	7.5±7.2*	13.8±10.1*	21.7±5.0*
Tongue chew	0.1±4.4	2.3±5.8	3.3±6.2	11.2±7.2*	17.3±7.1*	22.5±4.4*
Lip chew[†]	-1.1±2.9	0.9±5.0	0.9±5.5	6.1±9.5*	12.9±10.0*	22.0±5.3*

[†]Poor discrimination due to this task not being impaired in TD or mild CP; [α]Poor discrimination due to this task being 'impaired' in the TD group; *Significantly different from children with TD on logistic regression (binary outcomes) or linear regression (continuous outcomes); CP cerebral palsy; CI confidence interval; DDS Dysphagia Disorders Survey; GMFCS Gross Motor Function Classification System; NA Subtest as a whole was discriminative; PSAS Pre Speech Assessment Scale; SD Standard Deviation; SOMA Schedule for Oral Motor Assessment

Summary of Chapter 5

This paper presented novel data on the psychometrics of the SOMA, DDS and PSAS which have contributed to understanding the scores when applied to preschool children with CP. The inclusion of the third measure (PSAS), and a reference sample of children with typical development (TD), assisted in the interpretation of measure scores.

i. The findings suggest our previous prevalence estimate of 85% has likely included some children whose limitations to ingestion functions represent patterns associated with typical development. A more conservative estimate between 45% to 65% was proposed (45% representing the lowest modified prevalence estimate on the PSAS, and 65% representing the estimate generated from the web-based calculation).
ii. All 3 measures had similarly strong reproducibility (>85% for each of the measures overall). One measure should not, therefore, be prioritiesed over another based on this property.
iii. The DDS and PSAS were the pair of measures with the strongest agreement (in about 85% of cases), suggesting they are measuring a similar construct. Further work is needed to determine whether this construct is related to important health outcomes.
iv. OPD was present in about half of the sample with TD when using the published cut-points for each OPD measure. New OPD cut-points were proposed for each measure, using the mean (TD)±2SD.
v. The SOMA was calculated to have 100% specificity, the PSAS 63%, and the DDS 50%. The specificity of the PSAS and DDS improved when applying the modified cut-points. It is recommended that the modified cut-points should be applied when assessing children with CP aged 18 to 36 months.
vi. Applying the mofied cut-points to our sample yielded a lower overall OPD prevalence estimate, although it minimally impacted the prevalence in the nonambulatory group (GMFCS IV-V).
vii. Children with CP had consistently higher severity scores than children with TD. This supports the use of the DDS raw score as a valid measure of OPD severity.

Having established the reproducibility and validity of the OPD measures, and prevalence overall, it is important to begin to delineate the specific functional limitations to children's mealtimes. This was first explored with regards to the food and fluid textures children ingest, and the composition of children's diet with regards to gross motor function and OPD.

Chapter 6: Food and Fluid Textures Consumed by Preschool Children with Cerebral Palsy

Introduction to Chapter 6

This chapter presents the paper "Food and Fluid Texture Consumption in a Population-Based Cohort of Preschool Children with Cerebral Palsy: Relationship to Dietary Intake". It explored the food and fluid textures parents included in their child's diet and those that parents considered their child was able to eat. This was compared to a direct assessment of children's oral sensorimotor ability and safety on food/ fluid textures. Children's total energy intake and the texture composition of their diets were also analysed through 3-day weighed food records. By understanding each of these contributing factors in the context of dietary intake, we can construct better models for early screening and interventions.

Paper 5: Food and Fluid Texture Consumption in a Population-Based Cohort of Preschool Children with Cerebral Palsy: Relationship to Dietary Intake

This article was published in Developmental Medicine and Child Neurology, and has been cited twice (journal impact factor 3.292). Reproduced from Developmental Medicine and Child Neurology, Published online 23 March 2015 DOI: 10.1111/dmcn.12796 , Benfer, KA, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN, Food and Fluid Texture Consumption in a Population-Based Cohort of Preschool Children with Cerebral Palsy: Relationship to Dietary Intake, pages no. 1-8, Copyright 2015, © 2014 Mac Keith Press, with permission from John Wiley and Sons.

> Benfer KA, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Food and fluid texture consumption in a population-based cohort of preschool children with cerebral palsy. *Dev. Med. Child Neurol.* 2014. http://dx.doi.org/10.1111/dmcn.12796. Accessed May 15, 2015.

Data from this paper were also presented at the 7th Biennial Conference of the Australasian Academy of Cerebral Palsy and Developmental Medicine, March 2014, Hunter Valley, Australia; 67th Annual Meeting of the American Academy of Cerebral Palsy and Developmental Medicine, 16-19 October 2013, Milwaukee, United States; Speech Pathology Australia National Conference, June 2013, Gold Coast, Australia; and the European Academy of Childhood Disability, 8-11 June 2011, Rome, Italy.

Benfer KA, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Food textures habitually consumed by preschool-aged children with cerebral palsy: relationship to oropharyngeal dysphagia and gross motor functional skills. *Dev. Med. Child Neurol.* 2014;56(Supp 2):4. (Abstract)

Benfer K.A., Weir K.A., Bell K.L., Ware R.S., Davies P.S.W., Boyd R.N. Food textures habitually consumed by preschool aged children with cerebral palsy: relationship to oropharyngeal dysphagia and gross motor functional skills. *Dev. Med. Child Neurol.* 2013;55(Supp 3):32. (Abstract)

Weir KA, Benfer K, Bell K, Davies P, Robinson P, Ware R, Boyd RN. Oral feeding ability on food and fluid textures, and their relationship with gross motor skills in young children with cerebral palsy, *Dev. Med. Child Neurol.* 2011;53(Supp 5):14. (Abstract)

Food and fluid texture consumption in a population-based cohort of preschool children with cerebral palsy: relationship to dietary intake

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PUBLICATION DATA
Accepted for publication 23rd March 2015.
Published online

ABBREVIATIONS

CPFQ	Queensland Cerebral Palsy Child Feeding Questionnaire
DDS	Dysphagia Disorders Survey
OPD	Oropharyngeal Dysphagia
PEDI	Pediatric Evaluation of Disability Inventory

AIM To determine the texture constitution of children's diets and its relationship to oropharyngeal dysphagia (OPD), dietary intake, and gross motor function in young children with cerebral palsy (CP).

METHOD A cross-sectional, population-based cohort study comprising 99 young children with CP (65 males, 35 females) aged 18 to 36 months (mean age 27mo; Gross Motor Function Classification System [GMFCS] level I, n=45; II, n=13; III, n=14; IV, n=10; V, n=17). CP subtypes were classified as spastic unilateral (n=35), spastic bilateral (n=49), dyskinetic (n=5), and other (n=10), in accordance with the criteria of the Surveillance of Cerebral Palsy in Europe. Habitual dietary intake of food textures, energy, and water were determined from parent-completed 3-day weighed food records. Parent-reported feeding ability of food textures was reported on the Pediatric Evaluation of Disability Inventory and a feeding questionnaire. OPD was classified based on clinical feeding assessment using the Dysphagia Disorders Survey (rated by a certified assessor, KAB) and a subjective Swallowing Safety Recommendation (classified by a paediatric speech pathologist, KAB).

RESULTS Food/fluid textures were modified for 39% of children. Children with poorer gross motor function tended to receive a greater proportion of energy from fluids (GMFCS levels IV–V: β=0.9, p=0.002) in their diets and fewer chewable foods (level III: β=−0.7, p=0.03; levels IV–V: β=−1.8, p<0.001) compared to level I to II participants. Fluids represented a texture for which children frequently had OPD and the texture most frequently identified as unsafe (or recommended for instrumental assessment).

INTERPRETATION These findings indicate that swallowing safety, feeding efficiency, and energy/water intake should be considered when providing feeding recommendations for children with CP.

Cerebral palsy (CP) is a group of non-progressive motor disabilities[1] that can affect the range, strength, and coordination of motor control, including that required for eating, drinking, and controlling saliva. Oropharyngeal dysphagia (OPD) is common in approximately 85% of preschool children with CP,[2] and is used here to describe difficulty with oral movements necessary for efficient preparation and transport of the bolus, or pharyngeal movements which are important to swallow safely. OPD may limit the range of food/fluid textures children can safely and efficiently consume, which can lead to reduced dietary intake affecting nutritional status. It has been widely documented that children with CP are shorter and lighter compared to their peers.[3]

Food and fluid textures are classified using standardized terminology according to similarities in their physical properties (e.g. firmness and flow rate).[4] Modifications to food/fluid textures may be recommended to address mealtime safety or efficiency, or to encourage development of oral sensorimotor skills.[5] There is a general consensus that risk of aspiration (food or fluid entering the trachea below the vocal folds) is increased when consuming thin fluids.[6] Chewable foods may present a choking hazard if poorly masticated, and they are less efficiently consumed compared to purées in children with CP and those with typical development.[7] Foods typically introduced to children at a younger age, such as purées, are processed on the midline with a suckle feeding pattern, while more complex textures,

DOI: 10.1111/dmcn.12796

such as chewable foods, require tongue lateralization and separation of movement (of lip, tongue, and jaw function).[8]

There is a paucity of literature investigating the food and fluid textures in diets of preschool children with CP across the spectrum of motor severity. Studies have been skewed to school-aged children[9–11] and those with moderate to severe CP.[10–12] To our knowledge, no study has looked at habitual food/fluid texture consumption in children with CP. The Gross Motor Function Classification System (GMFCS) level and age of children varied between studies, as well as food texture terminology. This has resulted in variability in prevalence estimates, with the literature documenting modifications to solids in 25% to 64% of children with CP,[9–12] and fluid modification in 16%.[13] Earlier studies have had limited exploration of three major factors related to feeding ability on food/fluid textures, including gross motor function, OPD severity, and children's individual energy requirements. Children with CP with poorer gross motor function were reported to have poorer ability consuming textures,[14] and poorer gross motor function has also been associated with OPD.[2,9,11,12,15] Three studies have found an increased likelihood of texture modification in children with CP who have OPD.[9–11] Children with CP have also been found to have lower dietary energy intake than children with typical development,[16] which may be partially attributed to OPD[11] but may also reflect their lower energy requirements.[17]

This study aimed to explore the food/fluid textures consumed by preschool children with CP and the relationship with OPD severity and gross motor function. Understanding these factors will support better management decisions for feeding and nutrition in young children with CP, particularly when considering the safety and efficiency of textures consumed.

What this paper adds

- Food/fluid texture modifications present in 39% of preschool children with cerebral palsy, based on parent reporting.
- Children consumed equivalent amounts (grams), but energy intake decreased with poorer gross motor function.
- Children on average had 50% of intake as fluid, which was most commonly unsafely swallowed.
- Children with poorer gross motor function consumed less chewable items and more fluids compared to those with better gross motor function.

METHOD

This is a cross-sectional population-based study of young children with CP, conducted in Queensland, Australia from April 2009 to August 2013. It forms part of two longitudinal studies, Queensland CP Child: Motor Function and Brain Development (NHMRC 465128),[18] and Queensland CP Child: Growth, Nutrition and Physical Activity (NHMRC 569605).[19,20] The data reported from the Pediatric Evaluation of Disability Inventory (PEDI) represents a subset of the data reported by Weir et al.,[14] but includes only initial assessments and Queensland-born participants. It serves to determine the convergent validity of the PEDI with direct OPD assessment.

Participants

Participants were recruited through a range of settings from community to tertiary care. All children with a confirmed diagnosis of CP,[1] aged 18 to 36 months corrected age at initial assessment, and born in Queensland between 2006 and 2009 were invited to participate in the study. Children with neurodegenerative conditions were excluded.[18–20]

Measures

Two direct measures were used to clinically assess OPD, rated by a paediatric speech pathologist (KAB): the Dysphagia Disorders Survey – Pediatric (DDS) and a Swallowing Safety Recommendation. The DDS is a measure for screening signs of oral, pharyngeal, and oesophageal phase dysphagia.[21] The DDS Part 2 raw score indicates an individual's functional eating competency (maximum impairment=22). The DDS has been validated in 654 individuals with developmental disability aged 8 to 82 years, with strong agreement with blinded speech pathologist assessment (r=0.92) demonstrated in the final standardization study on 427 individuals (mean age 33y).[15] Interrater reliability has been shown to be strong in a study of 21 individuals and six speech pathologists (97% agreement).[21] The paediatric version was developed in a group of 166 children (range 2y 1mo–19y 1mo; mean 9y 4mo) with moderate to severe CP (GMFCS levels III–V) and intellectual disability.[15] The Swallowing Safety Recommendation was made by a speech pathologist for each food/fluid texture according to criteria for 'continue', 'supervise', 'refer', and 'exclude' (Appendix S1, online supporting information).

Parents reported on their child's ability and the inclusion of food/fluid textures using the PEDI and the Queensland Cerebral Palsy Child Feeding Questionnaire (CPFQ) based on their child's performance at home. The PEDI is a standardized measure of self-care, mobility, and social functioning, with demonstrated validity and strong inter- and intrarater reliability (ICC=0.95–0.99).[22] Items 1 to 4 indicate ability on 'puréed, blended, strained foods'; 'ground or lumpy foods'; 'cut up, chunky, diced foods'; and 'table foods'. Parents also reported on inclusion of purées, thick purées, lumpy/mashed foods, chewable foods, and tough chewable foods, and thin fluids and thickened fluids (levels 1–3, indicating level of thickness) in their child's diet using the CPFQ.[4] A parent-completed, 3-day weighed food record was used to measure habitual dietary intake (energy and water). Parents were instructed to record the amount of food and fluid consumed, and loss due to spillage.[23] This method is valid for assessing energy intake in preschool aged children with CP.[24]

The GMFCS classifies children's gross motor function according to five levels.[25] The age bands 'under 2 years' and '2- to 4-years' were used,[25] rated by two physiothera-

pists. Children's motor type (spastic, dyskinetic, and hypotonic) and distribution (unilateral, bilateral, and number of limbs) were classified according to the Surveillance of Cerebral Palsy in Europe.[26]

Procedures

Children attended the hospital for a videoed clinical feeding assessment[20] and gross motor assessment. Parents completed the 3-day weighed food record for 2 weekdays and 1 weekend day in the month after their appointment.[24] Nutritional analyses of the food records were completed by a dietician using Foodworks 7 dietary analysis software (Xyris Software [Australia] Pty Ltd, Kenmore Hills, Queensland, Australia). Foods and fluids on the 3-day food record were categorized by texture, consistent with the Australian standardized terms: purée, semi-solid, chewable, thin fluid, thick fluid.[4] The proportion of food/fluid textures habitually consumed was reported based on amount (grams [g]) and energy (kilojoules [kJ]). Energy and water intake were compared to the estimated average requirements reported in the Australian Nutrient Reference Values.[27]

Ethics

Ethics approval has been gained through the University of Queensland Medical Research Ethics Committee (2008002260), the Children's Health Services District Ethics Committee (HREC/08/QRCH/112), and at other regional and organizational ethics committees (see protocol paper for full list).[19] All families gave written informed consent to participate.

Statistical analyses

All data analyses were performed using Stata Statistical Software version 10.0 (StataCorp LP, College Station, TX, USA), with significance set at $p<0.01$. Demographic data were presented descriptively and compared to non-participants (using Fisher's exact test) and CP Register data (using χ^2 text for trends) for sex, GMFCS, and motor type (condition on available data only). GMFCS levels were combined into functionally similar groups to increase power for analyses (I–II, III, IV–V). The proportion of children with (1) each food/fluid texture excluded (CPFQ) and with (2) limited ability on each food (PEDI), and their relationship to GMFCS were explored using binomial logistic regression. For variables that predicted an outcome perfectly, exact conditional logistic regression has been used. The presence of a modified diet (indicated by a 'no' response to inclusion of any texture) and a child's inability on all table foods (indicated by 'no' response on 'able to eat all textures of table foods') were considered as binary variables in order to determine whether parent-reported inability of their child on all foods corresponded to parents modifying their child's diet. The concordance between these two parent-reported measures was calculated using percentage agreement. Similarly, the Swallowing Safety Recommendation was analysed as a binary variable, to differentiate those children who were safe on a texture (recommended to 'continue' or 'continue with supervision') from those considered unsafe ('refer for instrumental evaluation' and 'exclude'). The percentage agreement was also analysed between (1) parent-reported exclusion/inability on each texture, and (2) the directly assessed measures of OPD (OPD on the DDS [binary] and Swallowing Safety Recommendation – safe vs unsafe).

When investigating the association between GMFCS and textures consumed as a proportion of diet, we accounted for the composite nature of the purée, semi-solid, chewable, fluid, and tube variables by using the centred log-ratio transformation before regression modelling.[28,29] Models included (1) GMFCS category and (2) DDS raw score as the main effects and age as a covariable. To ensure all values were non-zero, 0.5% was added to each category and then rescaled before transformation.

The three textures assessed on the DDS (non-chewable, chewable, and fluid) were analysed for their association with (1) texture exclusion, (2) proportion of texture habitually consumed, and (3) Swallowing Safety Recommendation. Mean energy and water intake, and the proportion meeting estimated average requirements by GMFCS level were reported descriptively. The influence of factors (GMFCS and proportion of food textures habitually consumed) on children's energy intake was explored using linear regression. Adjusted models included the covariates of age and sex.

RESULTS

There were 178 eligible children referred, of which 132 children consented to participate in the Growth, Nutrition and Physical Activity study, with 99 completing three full days of the weighed food record. Of the children who declined participation, 18 participated in only the concurrent motor study (finding the burden of two studies too great), and 28 declined both studies (eight because of study burden, 13 because of family circumstances, two were non-English speaking, four resided interstate, and one passed away). Participants' age ranged from 17 to 37 months corrected age 2 years 3 months. Participant characteristics, and a comparison to non-participants and the Australian CP population,[30] are presented in Table I.

The proportion of children whose parents identified modified diets or limited ability on textures is shown in Table II. There were 39% of children with modified diets (on the CPFQ), and this proportion increased as gross motor function declined: levels I to II=19%, III=50% (adjusted OR=3.5, $p=0.06$), IV to V=78% (adjusted OR=31.9, $p<0.001$). Beyond the exclusion of tough chewables, only 23% had a modified diet. Based on parent report on the PEDI, 41% had limited ability in eating a full range of table foods. The number of children with limited ability increased as gross motor function declined (I–II=26%, III=36% [adjusted OR=1.3, $p=0.69$], IV–V=78% [adjusted OR=16.6, $p<0.001$]). There was evidence for

Table I: Characteristics of participants in the study

	Participants (%), *n*=99	Non-participants (%), *n*=79	Fisher's exact *p*-value: non-participants	Australian CP Register (%), *n*=2960	χ^2 (*p*-value): CP Register
Sex, males	66	61	0.58	57	3.4 (0.064)
GMFCS level			0.06		9.7 (0.045)
I	46	30		36	
II	13	17		25	
III	14	2		11	
IV	10	9		12	
V	17	13		14	
Unknown	0	29		4	
Primary motor type:		N/A	N/A		0.017[a]
Unilateral spasticity	35			33	
Bilateral spasticity	50			53	
Dyskinetic	5			6[a]	
Ataxic	2			5	
Hypotonic	8			2	
Unknown	0			1	
Motor distribution		N/A	N/A	N/A	N/A
One limb	2				
Two limbs	53				
Three limbs	9				
Four limbs	36				
Tube fed		N/A	N/A	N/A	N/A
Partial	10				
Complete	2				

Comparisons conditional on known data; [a]Fisher's exact test used due to <5 in cells. CP Register data taken from Australian Cerebral Palsy Register Group.[30] GMFCS, Gross Motor Function Classification System; N/A, data not available.

Table II: Parent-reported texture limitations on the Queensland Cerebral Palsy Child Feeding Questionnaire (CPFQ) and Pediatric Evaluation of Disability Inventory (PEDI), according to Gross Motor Function Classification System (GMFCS)

Texture excluded: CPFQ	% of GMFCS level (CI)	Crude OR (*p*)	Adjusted OR (*p*)	Limited ability on texture: PEDI	% of GMFCS level (CI)	Crude OR (*p*)	Adjusted OR (*p*)
GMFCS I–II (*n*=58)				GMFCS I–II			
Purée	3 (0–8)	Ref	Ref	Puréed, blended, strained	2 (0–5)	Ref	Ref
Semi-solid	7 (0–14)			Ground or lumpy	5 (0–11)		
Chewable	0 (0–6)			Cut up, chunky, diced	7 (0–14)		
Tough chewable	14 (5–23)			Table foods	26 (14–37)		
Thin fluid	0 (0–6)						
GMFCS III (*n*=14)				GMFCS III			
Purée	0 (0–23)	1.7 (1.00)[a]	1.0 (0.71)[b]	Puréed, blended, strained	0 (N/A)	4.1 (1.00)[a]	3.1 (1.00)[b]
Semi-solid	0 (0–23)	0.8 (0.83)[a]	0.6 (1.00)[b]	Ground or lumpy	0 (N/A)	1.1 (1.00)[a]	1.0 (1.00)[b]
Chewable	14 (0–34)	10.6 (0.07)[a]	8.9 (0.10)[b]	Cut up, chunky, diced	21 (0–44)	3.7 (0.12)	3.2 (0.17)[c]
Tough chewable	50 (23–78)	6.1 (0.006)	5.2 (0.014)[c]	Table foods	36 (9–62)	1.6 (0.46)	1.3 (0.69)[c]
Thin fluid	0 (0–23)	1.0 (NC)	1.0 (NC)				
GMFCS IV–V (*n*=27)				GMFCS IV–V			
Purée	15 (0–29)	4.8 (0.16)[a]	4.3 (0.25)[b]	Puréed, blended, strained	11 (0–23)	7.0 (0.19)[a]	4.7 (0.33)[b]
Semi-solid	44 (25–64)	10.4 (<0.001)[a]	10.7 (<0.001)[b]	Ground or lumpy	44 (25–64)	14.1 (0.001)[a]	14.4 (<0.001)[b]
Chewable	44 (25–64)	59.6 (<0.001)[a]	57.3 (<0.001)[b]	Cut up, chunky, diced	67 (48–85)	27.0 (<0.001)	33.4 (<0.001)[c]
Tough chewable	74 (57–91)	17.5 (<0.001)	26.8 (<0.001)[c]	Table foods	78 (62–94)	10.0 (<0.001)	16.6 (<0.001)[c]
Thin fluid	37 (18–56)	44.0 (<0.001)[a]	41.8 (<0.001)[b]				

[a]Cell value=0 for one or more groups, therefore analysed with exact conditional logistic regression; [b]adjusted for age only (not calculable for age and sex using Stata as memory exceeded for exact conditional logistic regression; [c]adjusted for age and sex. Note, exclusion/inability on each texture are not mutually exclusive (i.e. children may have more than one texture excluded/with inability), therefore total proportion with impairments per texture may not correspond to total with modification overall. OR, Odds ratio; CI, confidence interval; ref, reference group; N/A, no confidence intervals, as no children in this group; NC, not calculable as no children in reference or comparison groups.

an effect of age on the modification of the child's diet (OR=0.9, *p*=0.006) but no evidence for an effect on inability to consume a full range of table foods (OR 0.9, *p*=0.08). Four children (4%) were restricted to thickened fluids, all of whom were classified in GMFCS level V. Concordance between the CPFQ and PEDI was high for purées (98.0%), semi-solids (95.0%), chewables (88.9%), and tough chewables/table foods (86.7%).

The average daily habitual intake (taken orally or via tube, and not including quantities lost due to spillage) of

quantity of food/fluid (g), energy from food (kJ), and water content (g) is presented in Table III. This table also presents the proportion of children from combined GMFCS levels meeting estimated average requirements. Children's energy intake decreased with increasing GMFCS level, although the amount of food/fluid consumed and water intake was not significantly related to GMFCS level. Energy intake had a strong positive association with the proportion of chewables in children's diets (kJ: β=11.2, p=0.01; grams: β=16.7, p=0.03) and the proportion of fluids consumed (g: β=14.7, p<0.01). Water intake was strongly associated with consuming a lower proportion of chewable foods (kJ: β=−3.6, p<0.01; grams: β=−7.5, p<0.01).

The proportion of total average habitual energy intake from food/fluid textures by GMFCS level is shown in Figure 1 (see also Fig. S1, online supporting information, for proportion by weight in g). Children classified in GMFCS level III had a significantly lower proportion of overall energy from chewables compared to children in GMFCS levels I to II. Children in GMFCS levels IV to V and those who were tube fed had a significantly lower proportion of chewables and greater proportion of fluids in their diet compared to children in GMFCS levels I to II. There was evidence for an effect of age on the proportion of purées and chewables in children's diets, by GMFCS level. Children's score on the DDS significantly influenced the proportion of textures in children's diets for chewables and fluids, as shown in Figure S1. There was evidence for an effect of age on the percentage of purées and fluids habitually consumed when considering the influence of the DDS.

Children's OPD severity and safety on each texture, and the frequency of texture consumption, are presented in Table SI (online supporting information). Parents were found to include or report ability on food/fluid textures in their child's diet for which their child had OPD in 41% to 64% of children (see Table SI). However, the agreement between parent-reported inclusion/ability and clinician's Swallowing Safety Recommendation (continue or supervision only) was much higher (72–79% children, Table SI). Only three children who had food/fluid orally were assessed to be unsafe on all textures (based on the single clinical feeding assessment), and therefore recommended to have all textures excluded. The proportion of children who were recommended for exclusion of one or more textures was 7% of children on purées, 15% of those on chewables, and 9% for those drinking thin fluids. The recommendation for referral to instrumental assessment was made in 13% for purées, 3% for chewables, and 13% for thin fluids.

DISCUSSION

Modified food/fluid textures are a common feature of the diets of children with CP aged 18 to 36 months, reported by just under half of the parents. Modifications to diets also corresponded to parents reporting limitations in their child's ability on food textures, indicating parents are generally excluding foods/fluids for which they perceive their child has difficulty. For about half of the children, the modification was only exclusion of tough chewable foods. This may reflect milder OPD or typical family preferences for children of this age. Only children with moderate to severe CP were restricted to purées/semi-solids, representing a small percentage of the group overall (14%), although this constituted one-third of children classified in GMFCS levels III to V. This was consistent with two previous studies, finding about one-third of children (32–35%) were restricted to purées/semi-solids, with these studies only recruiting from GMFCS III to V.[10,31] In the present study, 10 children did not consume thin fluids (four drank thickened fluids, six via tube), which was less than the 16% consuming thickened fluids reported by Wil-

Table III: Average daily habitual intake of macronutrients and proportion meeting their estimated average requirement, by Gross Motor Function Classification System (GMFCS): orally and tube-fed children with cerebral palsy

	Mean (SD)	% meeting EAR	Crude β (p)	Adjusted β (p)[a]
Quantity (g)				
GMFCS I–II (n=58)	1078.1 (266.6)	N/A	Ref	Ref
GMFCS III (n=14)	1032.9 (490.2)	N/A	−45.9 (0.64)	−35.9 (0.72)
GMFCS IV–V (n=15)	1120.1 (430.7)	N/A	41.3 (0.67)	47.7 (0.62)
Tube fed (n=12)	1068.6 (207.2)	N/A	−10.2 (0.92)	−14.4 (0.90)
Energy (kJ)				
GMFCS I–II (n=58)	4310.1 (785.3)	53	Ref	Ref[b]
GMFCS III (n=14)	3941.7 (1611.5)	21	−368.4 (0.24)	−287.7 (0.35)
GMFCS IV–IV (n=15)	3709.7 (1136.7)	27	−600.5 (0.048)	−598.0 (0.049)
Tube fed (n=12)	3352.8 (1162.9)	17	−957.4 (0.004)	−1069.2 (0.003)
Water (g)[c]				
GMFCS I–II (n=58)	837.3 (256.7)	2	Ref	Ref
GMFCS III (n=14)	819.7 (413.1)	7	−17.6 (0.85)	−17.2 (0.86)
GMFCS IV–V (n=15)	929.8 (402.8)	13	92.5 (0.31)	102.7 (0.27)
Tube fed (n=12)	963.7 (334.3)	8	126.4 (0.21)	141.9 (0.20)

Relationship between dietary intake and GMFCS explored using linear regression. [a]Adjusted for age and sex; [b]Evidence for effect of age on model; [c]Proportion meeting 80% of water requirements GMFCS I–II=10%, III=7%, IV–V=20%, tube=8%. EAR, estimated average requirement; β, beta coefficient (linear regression); g, grams; N/A, no confidence intervals, as no children in this group; ref, reference group; kJ, kilojoules.

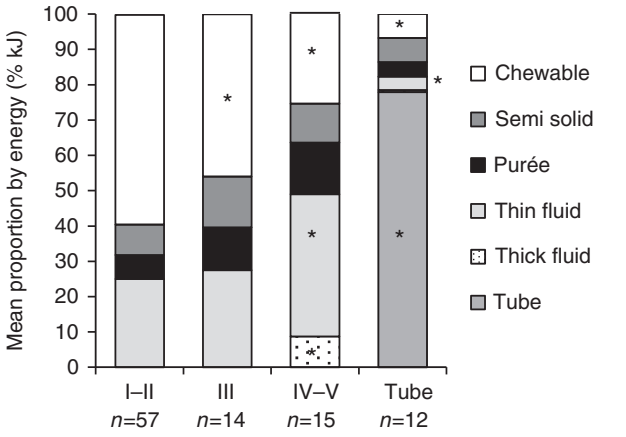

Figure 1: Diet constituency by texture according to Gross Motor Function Classification System (GMFCS) level in a population sample of preschool children with cerebral palsy. *indicates GMFCS levels for which the average proportion of texture intake differs significantly compared to GMFCS I to II. All *p*-values presented are adjusted for age, †indicates evidence of effect of age on model linear regression (using centred log-ratio transformation) for texture composition and GMFCS (base GMFCS level I–II) significant for: chewable† (III kJ β=−0.7, p=0.03; IV–V kJ β=−1.8, p<0.001; tube kJ β=−3.1, p<0.001), fluid (IV–V kJ β=0.9, p=0.002; tube kJ β=−2.0, p<0.001). kJ, kilojoules.

son et al.[13] Their study did not include data on children's motor severity, so differences in the motor severity between samples cannot be accounted for. The present study and our team's previous work on the same larger study[14] found that poorer gross motor function is associated with increased difficulty with food/fluid textures, and consequently increased modification to food/fluid textures. This specific relationship has not been reported on previously, but is consistent with the association reported between increased OPD severity and poorer gross motor function.[2,9,11,12,15]

The amount of food/fluid consumed by children with CP was on average consistent across the sample; however, the energy intake and texture constitution between GMFCS levels differed markedly. Regardless of GMFCS level, children tended to consume about a kg of food/fluid daily, but energy intake ranged from an average of 3084kJ for children fed by tubes to 4310kJ for children classified in GMFCS levels I to II. Children in GMFCS levels III to V and those with more severe OPD were found to consume a lower proportion of chewable foods and more fluids. About half of all children's diets were made up of fluids, increasing to almost two-thirds for children in GMFCS levels IV to V. Purées and fluids are likely to be more efficiently eaten by children with lower gross motor function, so despite these children receiving less energy from equivalent amounts of foods, these textures may still be a more efficient means of achieving adequate energy intake. Children consuming diets with less chewable foods may require an increased amount of food/fluid or have parents modify the energy density of easier to manage textures in order to achieve adequate energy intake.

Parents have been reported to under-detect OPD in their child with CP compared to direct assessment.[15] Our study found there was generally poor agreement (40–60%) between the parent-reported and direct OPD assessment (DDS); however, there was better agreement when considering the child's swallowing safety (70–80%). This indicates that parents may not consider specific oral phase impairments (e.g. limitations to biting or drinking from a cup) as 'limited ability' on that texture, but that they are accurately identifying many food/fluid textures for which their child may need referral or have excluded from their diet.

The relationship between the severity of OPD and swallowing safety on food/fluid textures, and the frequency of that mealtime risk (i.e. proportion of the texture in the child's diet) has not been discussed in the literature. Our results found that children with better gross motor function tended to have 'no to mild' OPD, and this translated into children generally being recommended as safe to continue with the texture, or possibly requiring supervision (mostly with chewable foods). The only exception was 10% of children from this group were recommended for referral to instrumental assessment for thin fluids, which reflects the greater complexity in managing this texture, particularly for young children. Children with moderate limitations in their gross motor function had only mild OPD with purées and fluids; however, they had moderate OPD on chewable foods. This group was mostly managing purées safely, but about half were recommended to receive supervision on thin fluids and chewables, and a small group were recommended for instrumental assessment. About one-quarter of their diets consisted of chewable foods and half of thin fluids, thus representing a group who may be a priority for feeding intervention. Finally, those with the poorest gross motor function had more severe OPD on all textures, with many recommended for either referral or exclusion (chewable foods most commonly).

Limitations

One of the largest limitations for feeding research remains the lack of a criterion standard measure of OPD, particularly one that validates the construct with nutritional status and safety. The use of the DDS as an objective direct assessment was considered the most appropriate standard against which to compare the parent-reported measures, in addition to a subjective Swallowing Safety Recommendation. A newly published classification system, the Eating and Drinking Ability Classification System,[32] shows promise to address such limitations for research in the field. This system is only valid for children as young as 3 years, so was not applicable in the current study.

The adequacy of children's energy and water intake was compared to estimated average requirements rather than individualized assessment of requirements. Previous work

from our team has found that children with poor gross motor function may have lower energy requirements than children with better gross motor function (often being smaller and less active).[17] Diminished growth of children with CP compared to their peers may begin in infancy and increases with age.[3] Children's nutritional status in early childhood may be attributed to inadequate intake during infancy,[16] although the aetiologies of poor growth in children with CP are multifactorial and unlikely to be related to OPD alone.[33] While we reported a large number of children not meeting estimated average requirements for dietary energy, it is important to interpret this finding considering these possible differences in current energy requirements.

Implications

To our knowledge, this is the first study to look at food and fluid intake in children with CP using directly assessed OPD, habitual texture intake, and a comparison to validated energy intake. The results have provided important data to inform management of children's dietary intake in this population. We found that while the two parent-reported measures performed consistently with each other, asking parents about inclusion/ability on a food/fluid texture in their child's diet is not a good marker of OPD overall, but was a reasonable indicator of safety. Training parents to detect safety concerns on food/fluid textures may be more clinically meaningful and effective than focusing on their identification of specific oromotor impairments. Using habitual texture intake from a 3-day weighed record gave more detailed information regarding texture consumption than previous methods based on inclusion of textures alone. This method may be useful for future research regarding the impact of children's intake on their health, as well as informing clinical recommendations.

This study also raises important points to consider when planning feeding and nutritional interventions for young children with CP. Children with poorer gross motor function were increasingly reliant on fluids for meeting their nutritional needs. As has been reported in previous studies, and supported by this study, children with CP may have more difficulty managing fluids and consequently have an increased safety risk. However, thin fluids may also be most efficiently consumed by these children, presenting a tension between safety and efficiency of dietary intake. Further exploration of the efficiency of intake and possible health consequences would help resolve this question.

Finally, the interaction between children's habitual texture consumption, OPD severity, and mealtime safety provides us with useful information to inform service planning based on children's GMFCS level. Children with moderate gross motor function (GMFCS level III) may represent a priority for oropharyngeal sensorimotor interventions to promote improved mealtimes and subsequent nutrition. Children with the lowest gross motor function would likely benefit from appropriate referrals and early nutritional interventions, including modifications to textures and alternative methods of nutrition (tube feeding). Swallowing safety, feeding efficiency, and energy/water intake should all be considered when providing feeding recommendations for children with CP, with ongoing monitoring to ensure that recommendations are implemented.

ACKNOWLEDGEMENTS

We would like to thank Physiotherapists Rachel Jordan (B Phty) and Chris Finn (B Phty) for data collection and gross motor ratings; and Dieticians Stina Oftedal (B Hlth Sc [Hons] Nutr & Diet) and Camilla Davenport (B Hlth Sc [Hons] Nutr & Diet) for data collection of feeding videos and diet analysis. This project was supported by the National Health and Medical Research Council Postgraduate Medical and Dental Scholarship (1018264 – KAB), Career Development Fellowship (APP1037220– RNB) and Project Grants (569605 and 465128). ANZTR Trial Registration Number: ACTRN12611000616976. The authors have stated that they had no interests that might be perceived as posing a conflict or bias.

SUPPORTING INFORMATION

The following additional material may be found online:

Figure S1. Diet constituency by texture according to (a) Gross Motor Function Classification System (GMFCS) level and (b) oropharyngeal dysphagia severity (Dysphagia Disorders Survey) in a population sample of preschool children with cerebral palsy**.**

Table SI. Relationship between directly assessed and parent-reported oropharyngeal dysphagia on food/fluid textures in preschool children with cerebral palsy.

Appendix A. Swallowing Safety Recommendation.

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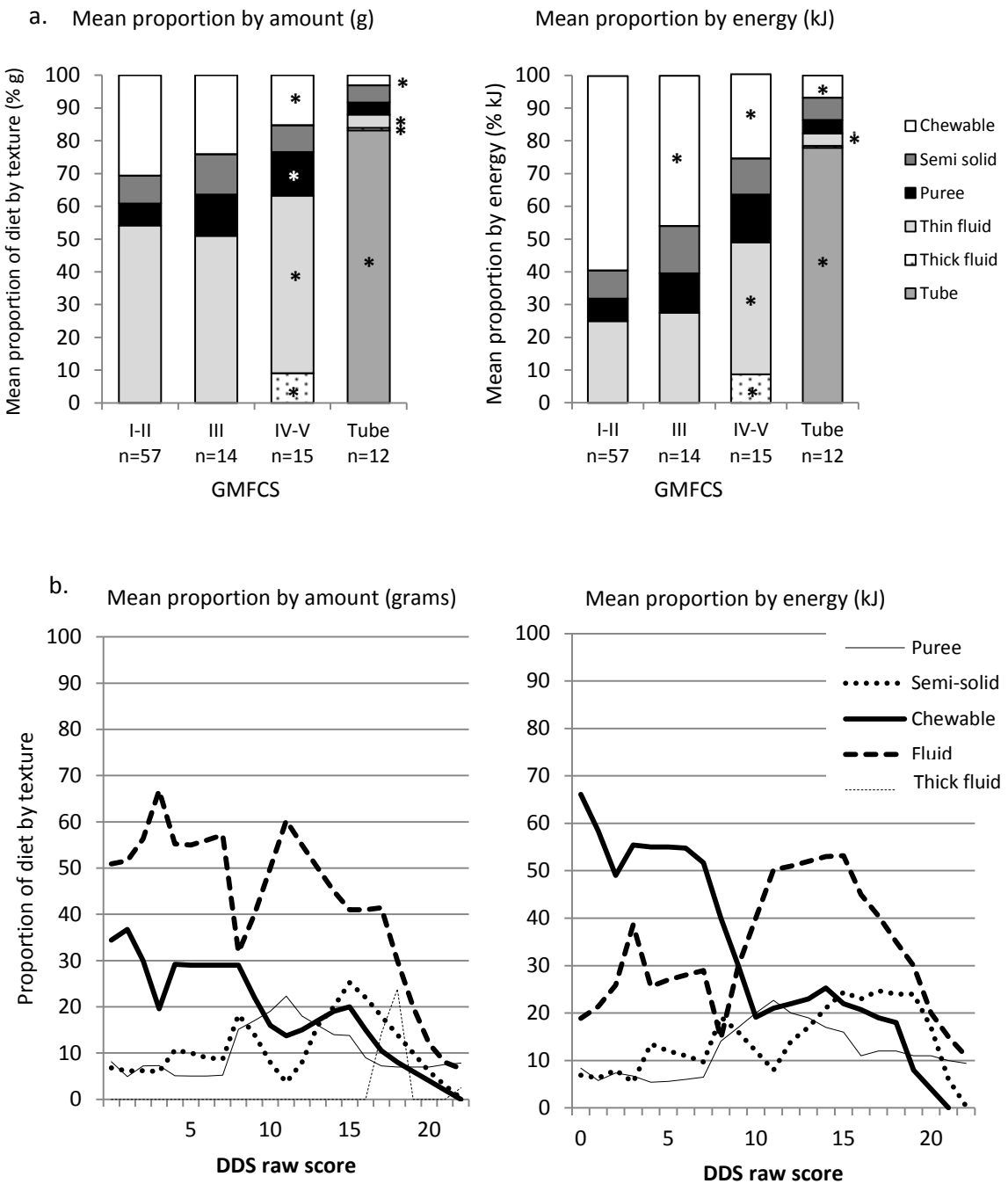
Appendix A: Swallowing Safety Recommendation

Continue: Child has oral sensorimotor & swallowing skills to safely and efficiently consume the texture with no need for supervision. No pharyngeal signs, but may have single cough on thin fluid.

Supervise: Supervision required (due to oral sensorimotor/swallowing skills or behavioural feeding skills), or requires significant mealtime modifications, or skilled mealtime assistance.

Referral: Refer for instrumental assessment. Referral may be indicated due to poor initiation of pharyngeal phase, a very disorganised oral phase, impaired motoric performance, multiple swallows/ poor laryngeal excursion, or clinical signs of aspiration (such as coughing, wet breathing, wet vocalisations, fremitus etc).

Exclude: Multiple clinical signs of pharyngeal phase impairment suggestive of oropharyngeal aspiration and placing child at risk of respiratory compromise. Or, oral phase skills on chewable food are very disorganised or impaired motoric performance placing child at risk of choking. If child is given extremely small pieces of chewable food with specific placement, this constitutes ‘exclusion’ of chewable foods.



Supplementary 1: Diet constituency by texture according to (a) gross motor function (GMFCS) and (b) oropharyngeal dysphagia severity (Dysphagia Disorders Survey) in a population sample of preschool children with cerebral palsy

Key: * indicates GMFCS levels for which the average proportion of texture intake differs significantly compared to GMFCS I-II. All p values presented are adjusted for age, [†]evidence of effect of age on model. Fluid analysed as combined variable (thin and thick fluids) (a) Linear regression (using centred log-ratio transformation) for texture composition and GMFCS (base GMFCS I-II) significant for: puree[†] (IV-V g β=0.7, p=0.01), chewable[†] (III kJ β=-0.7, p=0.03; IV-V g β=-1.4, p<0.001; IV-V kJ β= -1.8, p<0.001; tube g β=-2.6, p<0.001; tube kJ β=-3.1, p<0.001), fluid (IV-V kJ β=0.9, p=0.002; tube g β=-2.6, p<0.001; tube kJ β=-2.0, p<0.001); (b) Linear regression (using centred log-ratio transformation) for texture composition and Dysphagia Disorders Survey: significant for: chewable (kJ β=-0.2, p<0.001; g β=-0.1, p<0.001); fluid[†] (kJ β=-0.04, p=0.049; g β-0.1, p<0.001); Figure only shows points with >3 children; DDS Dysphagia Disorders Survey; GMFCS Gross Motor Function Classification System; kJ kilojoules

Summary of Chapter 6

The findings from this study suggest that swallowing safety, feeding efficiency, and energy/ water intake should all be considered when providing feeding recommendations for children with CP. More specifically, there were a number of important findings:

i. Modified diets were common in about half of the children with CP, and texture modification/ inability increased with increasing GMFCS level. Only children from GMFCS III-V were restricted to purees/ semi-solids.
ii. Exclusion of textures was closely related to parents' perception of their child's inability on the texture. This suggests parents are generally excluding textures for which they perceive their child has difficulty.
iii. Parents agreed with the direct assessment of OPD (on the DDS) in only 40% to 60% of cases, however this agreement was much better when considering directly assessed safety on the texture (70% to 80% agreement). Parents may not consider specific oral phase impairments (such as limitations to biting or drinking from a cup) as *inability* on that texture, but were often able to identify textures for which their child had safety concerns.
iv. The amount of food/ fluid children consumed was equivalent between gross motor levels, however the energy intake and texture composition of their diets differed markedly. About half of all children's diets were fluids, and this proportion was greater in children with nonambulatory CP (GMFCS IV-V). These children also ingested a lower proportion of chewable foods compared to ambulatory children with CP (GMFCS I-II).
v. The energy density of more efficiently managed textures should be modified so children can maintain adequate energy intake from smaller volumes of food.

This chapter has provided an understanding of the modification to food/ fluid textures in the diets of children with CP. Impairments to ingestion functions of the oral phase may influence a child's ability to safely and efficiently manage food and fluid textures. Thus, an increased understanding of these specific impairments may assist in understanding some of the underlying factors influencing children's diet modifications.

Chapter 7: Oral Phase Impairments in Preschool Children with Cerebral Palsy

Introduction to Chapter 7

This chapter focuses on the specific oral phase impairments associated with each classification level of gross motor function in preschool children with CP. It presents this by way of a published article, “Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy: Oral Phase Impairments”. This paper presents data on impairments to ingestion functions of the oral phase based on 3 measures – the SOMA, DDS and PSAS – and how oral phase impairments relate to mealtime duration, frequency and efficiency. These data were strengthened through the inclusion of a sample of children with TD, to which children with CP were compared.

Paper 6: Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy: Oral Phase Impairments

This article was published in Research in Developmental Disabilities (journal impact factor 2.735). Reprinted from Research in Developmental Disabilities, Vol 35, Benfer, K. A., Weir, K. A., Bell, K. L., Ware, R. S., Davies, P. S. W., & Boyd, R. N., Oropharyngeal dysphagia in preschool children with cerebral palsy: oral phase impairments, pages no. 3469-3481, Copyright 2014, with permission from Elsevier.

Benfer KA, Weir KA, Bell KL, Ware RS, Davies PSW, Boyd RN. Oropharyngeal dysphagia in preschool children with cerebral palsy: oral phase impairments. *Res. Dev. Disabil.* 2014;35:3469-3481.

Data from this paper were also presented as free papers at the 7th Biennial Conference of the Australasian Academy of Cerebral Palsy and Developmental Medicine, March 2014 and the 67th Annual Meeting of the American Academy of Cerebral Palsy and Developmental Medicine, 16-19 October 2013, Milwaukee, United States.

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Research in Developmental Disabilities 35 (2014) 3469–3481

Contents lists available at ScienceDirect

Research in Developmental Disabilities

Oropharyngeal dysphagia in preschool children with cerebral palsy: Oral phase impairments☆

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ARTICLE INFO

Article history:
Received 24 July 2014
Accepted 20 August 2014
Available online

Keywords:
Orpharyngeal dysphagia
Deglutition disorders
Oral phase impairment
Cerebral palsy
Children

ABSTRACT

Purpose: This study aimed to document the prevalence and patterns of oral phase oropharyngeal dysphagia (OPD) in preschool children with cerebral palsy (CP), and its association with mealtime duration, frequency and efficiency.
Methods: Cross-sectional population-based cohort study of 130 children diagnosed with CP at 18–36 months ca (mean = 27.4 months, 81 males) and 40 children with typical development (mean = 26.2, 18 males). Functional abilities of children with CP were representative of a population sample (GMFCS I = 57, II = 15, III = 23, IV = 12, V = 23). Oral phase impairment was rated from video using the Dyspahgia Disorders Survey, Schedule for Oral Motor Impairment, and Pre-Speech Assessment Scale. Parent-report was collected on a feeding questionnaire. Mealtime frequency, duration and efficiency were calculated from a three day weighed food record completed by parents. Gross motor function was classified using the Gross Motor Function Classification System (GMFCS).
Results: Overall, 93.8% of children had directly assessed oral phase impairments during eating or drinking, or in controlling saliva (78.5% with modified cut-points). Directly assessed oral phase impairments were associated with declining gross motor function, with children from GMFCS I having a 2-fold increased likelihood of oral phase impairment compared to the children with TD (OR = 2.0, $p = 0.18$), and all children from GMFCS II–V having oral phase impairments. Difficulty biting (70%), cleaning behaviours (70%) and chewing (65%) were the most common impairments on solids, and difficulty sipping from a cup (60%) for fluids. OPD severity and GMFCS were not related to mealtime frequency, duration or efficiency, although children on partial tube feeds had significantly reduced mealtime efficiency.
Conclusions: Oral phase impairments were common in preschool children with CP, with severity increasing stepwise with declining gross motor function. The prevalence and severity of oral phase impairments were significantly greater for most tasks when compared to children with typical development, even for those with mild CP. Children who were partially tube fed had significantly lower feeding efficiency, so this could be a useful

☆ ANZTR Trial Registration Number: ACTRN12611000616976.

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http://dx.doi.org/10.1016/j.ridd.2014.08.029

early indicator of children needing supplementation to their nutrition (through increasing energy density of foods/fluids, or tube feeds).

1. Introduction

Oropharyngeal dysphagia (OPD), or impaired feeding, has been frequently cited in the literature as an important factor influencing growth, nutritional status and respiratory health in children with cerebral palsy (CP) (Calis et al., 2008). Oropharyngeal dysphagia is common in approximately 85% of preschool children with CP (Benfer et al., 2013), largely related to the motor and sensory impairments associated with the diagnosis. Cerebral palsy is a group of non-progressive motor disabilities (Smithers-Sheedy et al., 2013) which can impact on the range, strength and coordination of motor control. The process of eating and drinking is commonly delineated into a number of interrelated phases, including the oral-preparatory, oral (propulsive), and/or pharyngeal phases (Matsuo & Palmer, 2008)

The oral-preparatory and oral propulsive phases (here-in described jointly as the 'oral phase') involve the child alerting to the bolus, receiving the bolus (through stripping a spoon, biting, or sipping from a bottle/cup), closure of the lips and nasopharynx to maintain the food/fluid in the mouth, moving the bolus in the mouth to prepare it for swallowing (including mastication), and propulsion of the bolus posteriorly for the initiation of the pharyngeal phase (Matsuo & Palmer, 2008). Impairments of the oral phase tend to receive the most emphasis in the feeding literature and in clinical assessments, in part due to the fact that they are more observable. The oral phase is important as it may impact on the efficiency of intake (e.g. increased anterior loss of food or fluids, increased oral transit time), which may lead to poor growth and nutrition. An impaired oral phase can also result in premature spillage of the bolus into the pharynx before the swallow has been initiated, piecemeal deglutition (the bolus being divided into multiple parts), and oral residue post-swallow, which can all impact on the safety of the mealtime.

A number of studies have explored oral phase impairments in children with CP, finding these impairments to be prevalent in between 68 and 95.4% of children (Field, Garland, & Williams, 2003; Gisel, Applegate-Ferrante, Benson, & Bosma, 1996; Gisel, Alphonce, & Ramsay, 2000; Kim, Han, Song, Oh, & Chung, 2013; Love, Hagerman, & Taimi, 1980; Mirrett, Riski, Glascott, & Johnson, 1994; Ortega, Ciamponi, Mendes, & Santos, 2009; Reilly & Skuse, 1992; Reilly, Skuse, & Poblete, 1996; Rogers, Arvedson, Buck, Smart, & Msall, 1994; Selley et al., 2001; Yilmaz, Basar, & Gisel, 2004; Yokochi, 1997). The variability in these estimates is related to participants' characteristics (in particular their age and gross motor severity), and the oral phase items/tasks assessed. Analysis of oral phase impairments in the literature has been based on a number of approaches; using a systems based approach (e.g. impairments to lips, tongue, jaw), analysis of ingestion functions (e.g. spoon feeding, biting, chewing, clearing), or documenting the specific oral sensorimotor impairments (e.g. prolonged oral transit time, impaired lip closure, use of extension-retraction tongue pattern). The most commonly cited impairments in individual studies included poor response to anticipation (Selley et al., 2001), difficulty stripping the spoon (Reilly & Skuse, 1992), poor lip closure (Ortega et al., 2009; Reilly & Skuse, 1992), difficulty drinking from a straw (Love et al., 1980; Ortega et al., 2009) and cup (Gisel et al., 2000), use of extension-retraction tongue patterns (Reilly & Skuse, 1992), difficulty chewing (Gisel et al., 1996, 2000; Ortega et al., 2009; Yilmaz et al., 2004), inadequate bolus formation (Kim et al., 2013), piecemeal deglutition (Kim et al., 2013) and oral residue (Kim et al., 2013). Many of the studies did not account for the gross motor function of the participants, therefore it is difficult to synthesise these data to characterise a picture of the patterns we would expect in the different functional levels.

The current study aimed to document the overall prevalence of oral phase OPD (overall and specific ingestion functions) in children with CP aged 18–36 months, as well as its association with mealtime frequency, duration and efficiency. Oral phase patterns were described with reference to a typically developing (TD) reference sample, and the child's gross motor function (on the Gross Motor Function Classification System, GMFCS), in order to understand how the specific oral phase impairments vary in children with this heterogeneous diagnosis. A better understanding of the range of functional impairments will assist in understanding which ingestion functions may be most important to consider for various health outcomes (such as growth and respiratory health), and which children may be most successfully targeted for therapy.

2. Materials and methods

This is a cross-sectional population-based study of preschool aged children with CP, conducted in Queensland, Australia between April 2009 and March 2013. It is part of two concurrent longitudinal studies exploring brain structure and motor function (National Health and Medical Research Council (NHMRC) 465128) and the relationship between growth, nutrition and physical activity (GNPA, NHMRC 569605) in children with CP. The design of the larger studies (Bell et al., 2010; Boyd et al., 2013) and current study (Benfer, Weir et al., 2012) have been described previously. Ethics approval has been gained through the University of Queensland Medical Research Ethics Committee (2008002260), the Children's Health Services District Ethics Committee (HREC/08/QRCH/112), and at other regional and organisational ethics committees (see protocol papers for full lists). All parents or guardians gave written informed consent to participate.

2.1. Participants

Children with a confirmed diagnosis of CP, aged 18–36 months corrected age at the time of initial assessment, and born in Queensland between 2006 and 2009, were invited to participate in the study in the CP sample. Children with neurodegenerative conditions were excluded from the study. An additional 40 children aged 18–36 months with typical development (term births >37 weeks, no diagnosis which required neonatal admission or ongoing medical or allied health treatment, and not on regular medication) were recruited through convenience sampling as a reference group.

2.2. Measures

Four direct measures of oral phase impairment were selected based on the results of a comprehensive systematic review of their psychometric properties and clinical utility (Benfer, Weir et al., 2012; Benfer, Weir, & Boyd, 2012), and further testing of their psychometrics (Benfer, 2014 [unpublished work]). Measures selected were the Schedule for Oral Motor Assessment (SOMA) (Reilly, Skuse, & Wolke, 2000), Dysphagia Disorders Survey (DDS) (Sheppard, 2003), Pre-Speech Assessment Scale (PSAS) (Morris, 1982), and the Thomas-Stonell and Greenberg Saliva Severity Scale (Thomas-Stonell & Greenberg, 1988). Prevalence data for each of these measures were presented based on their standard scoring and also modified cut-points developed in our previous work (Benfer et al., 2014 [unpublished work]).

The SOMA is a discriminative measure which identifies oral motor dysfunction in children according to skills that are typically mastered from 8 to 24 months (Reilly et al., 2000). It categorises oral motor dysfunction based on cut-scores for seven oral motor challenge categories (puree, semi-solid, solid, cracker, bottle, trainer cup, cup) (Reilly et al., 2000). The SOMA is predominantly a test of oral phase dysfunction; however, some items pertain to the pharyngeal phase.

The DDS is an evaluative measure for screening signs of oral, pharyngeal and oesophageal phase dysphagia in children and adults with a developmental disability (Sheppard, 2003). Part 2 was used to provide a raw score to indicate the child's functional eating competency (maximum impairment raw score of 22) and this subtest has been used previously as a measure of OPD (Calis et al., 2008). Items 8–12 (orientation, reception, containment, oral transport and chewing) are specifically oral phase tasks.

The PSAS is an evaluative measure that examines 27 pre-speech feeding behaviour performance areas related to sucking, swallowing, biting, chewing, respiration–phonation and sound play (Morris, 1982). Each subtest is scored on an ordinal abnormality scale, and a developmental scale (with age norms), to provide a double score overall. The sucking and biting/chewing subtests focus on the oral phase.

The Thomas-Stonell and Greenberg Saliva Severity Scale is a semi-quantitative assessment of drooling severity (one to five point scale of severity (no drooling to profuse drooling), and frequency (no drooling to continuous drooling)) based on observations of anterior saliva loss (Thomas-Stonell & Greenberg, 1988).

Parents reported on their child's OPD using the Queensland CP Child Feeding Questionnaire (CPFQ). Parents reported on the "severity of eating or drinking difficulties" for their child using two ten-centimetre visual analogue scales (VAS), with 0 being "no problems" and 10 being "major problems". A VAS score of greater than 0 was used to classify parent-reported oral phase impairments. They also reported on the presence of three specific oral phase impairments (difficulty drinking from spout cup or cup, difficulty moving food to the back of his/her mouth when eating from a spoon, and difficulty biting or chewing food).

The total daily feeding time was calculated from times recorded on a three day weighed diet record completed by parents at home (Bell et al., 2010). Nutritional analysis of records was completed by a dietician using Foodworks 7 dietary analysis software (Xyris Software (Australia) Pty Ltd.). Children with mealtime duration data recorded for a full day were reported as 'actual' time (daily average). 'Actual' mealtime efficiency (kilojoules per minute and grams per minute) was calculated only on data available (i.e. those energy and amounts with times recorded against them). Records with less than half of the times missing from food items had the missing times estimated based on an averaged time from their own record for consuming an equivalent texture (if greater than half were missing, this record was excluded from the time analysis). If there was no time recorded for any equivalent texture, estimated values could not be calculated, and therefore records excluded. This study defined a 'meal' as a distinct intake of energy which could be made up of food, drinks or both, with less than 5 min between foods.

Children's gross motor function is classified according to five levels on the GMFCS (Palisano et al., 1997). The <2 years and 2–4 year scales were used in this study (Palisano et al., 1997). The motor type (spastic, dyskinetic and hypotonic) and distribution (unilateral, bilateral, and number of limbs) were classified according to the Surveillance of CP in Europe (Sanger, Delgado, Gaebler-Spira, Hallett, & Mink, 2003).

2.3. Procedures

Children attended the hospital for mealtime and gross motor assessment. Mealtimes were videoed for later rating, as recommended in the SOMA administration manual, with children well positioned in their typical mealtime seating. Three standardised presentations of four textures (puree, lumpy, chewable and fluid) were presented by the primary carer, using their regular utensils (Reilly et al., 2000). Following these standard presentations, the child was allowed to complete the snack as usual. All gross motor ratings were conducted by two trained physiotherapists. Parents completed the weighed food records on two weekdays and one weekend day in the month following their appointment (Bell et al., 2010).

3. Calculations

All data analyses were performed using Stata 10.0 (Statacorp 2007), with significance set at $p < 0.05$ and 95% confidence intervals (CI) reported. Demographic data were presented with descriptive statistics. The association between overall and specific oral phase impairments and gross motor attainment (GMFCS) was explored using binomial logistic regression, with children with TD as the comparison group. Severity of OPD was reported as mean of the scores, and also converted into a scaled score from 0 to 10 to allow comparison between measures. Agreement between direct assessment and parent report were calculated based on percentage agreement (near perfect agreement for continuous outcomes ± 1), and kappa statistics (binary outcomes) or Intra-class Correlation Coefficients (ICC, continuous outcomes). Bias in the agreement between direct and parent-report was determined with mean of differences. The mean frequency, duration and efficiency (kJ and grams per minute) of mealtimes were reported according to GMFCS level (with children with complete tube feeding excluded, and those with partial tube feeding analysed as a separate sub-group). The influence of OPD severity and gross motor function was explored in relation to these four outcomes, using linear regression.

4. Results

There were 178 eligible children referred to the study, of which a total of 132 children consented to participate in the GNPA study, with 130 completing the mealtime assessment (Supplementary Information 1). Of the children who declined participation, 18 participated in only the concurrent Qld CP Child Motor Function and Brain Development Study (finding the burden of two studies too great), and 28 declined both studies (eight due to study burden, 13 due to family circumstances, two were non-english speaking, four resided interstate, and one passed away). Participants' ages ranged from 17 to 37 months corrected age at the time of assessment (mean = 27.4 months, SD = 5.3). The sample characteristics are presented in Table 1, and are representative of the population of Australian children with CP (Benfer et al., 2013).

4.1. Missing data

All children had an OPD classification for the direct assessments (DDS, SOMA, PSAS), with 122 children having a total score on the DDS. Four parents did not complete the feeding questionnaire. There were 110 children whose parents had completed one or more days on the food record, and 93 with sufficient data to estimate mealtime duration.

4.2. Prevalence and severity of oral phase impairments

Overall, 122 children with CP (93.8%) had directly assessed oral phase impairments during eating or drinking (based on the SOMA, DDS, and PSAS sucking and bite/chew subtests) or in controlling saliva. There were 66 children (50.8%) who had impaired saliva control, and only one of these children who did not have co-occurring oral phase impairments during eating/drinking. Directly assessed oral phase impairments were associated with poorer gross motor function, with children from GMFCS I having twice the odds of oral phase impairment compared to the children with TD (OR = 2.0, CI = 0.7–5.8, $p = 0.18$), and all children from GMFCS II–V having oral phase impairments. Based on the modified cut-points developed by our team (Benfer et al., 2014 [unpublished work]), this estimate for oral phase impairment is 78.5%, four of whom had only impaired saliva control.

Parents reported impairments to solids or fluids in 79.2% of their children (based on the VAS or impaired saliva control). There were 60.5% of children whose parents reported they had impaired saliva control, of whom 25 children who were not reported to have oral phase impairments in eating/drinking. While there was an overall trend for increasing prevalence of oral phase impairments with poorer gross motor function (GMFCS I = 71.9%, II = 66.7%, III = 78.3%, IV = 91.7%, V = 100.0%), parent-reported impairments were not found to be associated significantly with gross motor function (using GMFCS I as the comparison group).

Children's severity of OPD on solids was 3.2 on the scaled scoring (0–10) based on each of the DDS, PSAS and parent report (VAS). There was more variability between measures for severity on fluids, ranging from 1.1 on the SOMA and 3.3 on the DDS (Table 2). Children's mean OPD severity on the DDS increased stepwise as gross motor function declined, from 0.8 in children with TD to 19.1 in GMFCS V (Fig. 1) (solid scaled score TD = 0.1, GMFCS V = 8.8; Fluid scaled score TD = 0.8, GMFCS V = 8.6). Scores were significantly higher for all GMFCS levels when compared to the children with TD (correlation coefficient $r = 0.89$, $p < 0.01$).

4.3. Agreement between directly assessed and parent-reported oral phase impairments

The agreement (close or perfect) between directly assessed and parent-reported OPD severity was between 48 and 58% for solids and 39.4–62.8% for fluids, with ICCs ranging from 0.03 to 0.64 (Table 2 and Fig. 2). Parents did not consistently over or under estimate children's feeding severity (almost no bias (mean of differences <1.0) for the DDS and PSAS, and parents slightly over-reporting when compared to the SOMA), as shown in Fig. 2. Agreement on specific oral tasks was best for difficulty on cup/trainer cup according to the SOMA when compared to parent report (80.0%), and poorest for this same task compared to the PSAS (31.8%) (Table 2).

Table 1
Characteristics of participants in the Oropharyngeal Dysphagia Study.

	Participants, *n* (%)
Gender, males	81 (62.3)
GMFCS level	
I	57 (44.2)
II	15 (11.6)
III	23 (17.8)
IV	12 (9.3)
V	23 (17.7)
Primary motor type	
Unilateral spasticity	41 (31.5)
Bilateral spasticity	72 (55.4)
Dystonia	2 (1.5)
Ataxia	2 (1.5)
Hypotonia	9 (6.9)
Athetoid	4 (3.1)
Motor distribution	
One limb	2 (1.6)
Two limbs	67 (51.5)
Three limbs	13 (10.0)
Four limbs	48 (36.9)
Tube fed	
Partial	11 (8.4)
Complete	5 (3.9)
Primary utensil: bottle	
I	3 (5.3)
II	0 (0.0)
III	5 (21.7)
IV	6 (50.0)
V	3 (13.6)
Primary utensil: trainer cup	
I	29 (50.9)
II	8 (53.3)
III	13 (56.5)
IV	3 (25.0)
V	5 (22.7)
Primary utensil: cup	
I	25 (43.9)
II	7 (46.7)
III	4 (17.4)
IV	3 (25.0)
V	2 (9.1)

Key: GMFCS, Gross Motor Function Classification System; SD, standard deviation.

4.4. Specific oral phase impairments

The range of specific oral phase impairments for children with CP and TD, as identified on the three direct OPD measures, are presented in Table 3 (solids) and Table 4 (fluids). Only one child with TD used a bottle for daytime fluids, eight used trainer cups and 38 a standard cup. Of the children with CP, 33 were observed using a bottle, 77 trainer cup (including pop-top and straw), and 66 with cups (Table 1). There were more children with CP who used bottles (OR = 6.0, $p = 0.09$) and trainer cups (OR = 7.6, $p < 0.01$) compared to children with TD, and the proportion not using cups increased with GMFCS level (TD = 12.5%, I = 57.1%, II = 53.3%, III = 81.8%, IV = 75.0%, V = 90.1%). No children from the TD group refused fluids in the mealtime assessment, compared to eight children (6.2%) in the CP group.

4.5. Mealtime frequency, duration and efficiency

The feeding frequency, duration and efficiency by gross motor level are shown in Table 5. The average daily feeding duration ranged from 27 min to 6 h 49 min, with 24.7% of children having feeding times greater than 3 hours per day (based on estimations in the absence of full day data). Only feeding efficiency (kJ and grams) was significantly associated with children who had partial tube feeds, compared to those in GMFCS I. OPD severity (on the DDS) was not related to mealtime frequency, duration or efficiency.

Table 2
Severity of oropharyngeal dysphagia, and agreement between directly assessed and parent-reported oral phase impairments in young children with cerebral palsy.

	Parent-reported (CPFQ)	Direct assessment			Agreement[a] between direct assessment and parent-report
		DDS	SOMA	PSAS	
Severity of eating problem					
Mean score (CI)	3.2 (2.6–3.9)	4.8 (3.9–5.7)[e]	5.9 (3.6–8.2)[f]	39.6 (31.4–37.7)[g]	–
Scaled mean (CI)	3.2 (2.6–3.9)	3.2 (2.6–3.8)	1.5 (0.9–2.5)	3.2 (2.5–3.8)	DDS: 54.0%, ICC = 0.62, $p < 0.01$ SOMA: 58.1%, ICC = 0.30, $p = 0.02$ PSAS: 47.8%, ICC = 0.64, $p < 0.01$
Severity of drinking problem					
Mean score (CI)	2.5 (1.9–3.2)	2.3 (1.9–2.8)[e]	NA	8.5 (5.4–11.6)[g]	–
Scaled mean (CI)	2.5 (1.9–3.2)	3.3 (2.7–3.9)	1.1 (0.7–1.4)[f]	1.9 (1.3–2.5)	DDS: 39.4%, ICC = 0.60, $p < 0.01$ SOMA: 61.1%, ICC = 0.03, $p = 0.35$ PSAS: 62.8%, ICC = 0.60, $p < 0.01$
Difficulty on cup/t-cup[b] % (CI)	19.2 (12.4–26.1)	68.6 (60.3–76.9)	Cup: 21.2 (11.1–31.3) t-cup: 27.3 (17.1–37.5)	41.1 (31.6–50.6)	DDS: 48.4%, kappa = 0.16, $p < 0.01$ SOMA: 80.0%, kappa = 0.34, $p < 0.01$ PSAS: 31.8%, kappa = 0.03, $p = 0.18$
Difficulty moving food to back of mouth from spoon[c] % (CI)	13.1 (7.2–19.0)	28.6 (20.3–36.8)	NA	NA	DDS: 76.5%, kappa = 0.30, $p < 0.01$
Difficulty biting/chewing[d] % (CI)	26.9 (19.2–34.7)	62.1 (53.1–71.0)	47.7 (38.2–57.2)	63.1 (54.4–71.8)	DDS: 54.3%, kappa = 0.19, $p < 0.01$ SOMA: 59.6%, kappa = 0.17, $p = 0.02$ PSAS: 50.8%, kappa = 0.12, $p = 0.04$

[a] Agreement to 1 point either side of perfect agreement for continuous variable.
[b] Proportion based on DDS impairment to fluid subtest, SOMA impairment to trainer cup or cup oral motor challenge category, PSAS impairment to 'sucking from cup'.
[c] Proportion based on DDS item 11 oral transit for non-chewable.
[d] Proportion bsed on DDS impairment to chewable reception, SOMA impairment to 'bite' items, PSAS impairment to biting/chewing subtest
[e] Eating problem: sum of non-chewable and chewable subtests (out of 15), $n = 125$, drinking problem: fluid subtest (out of 7).
[f] Eating problem: puree + semi-solid + cracker raw score (out of 39), $n = 65$, drinking problem: fluid raw score converted to 0–10 scale using sum of items on most commonly used fluid utensil, $n = 118$.
[g] Mean months delayed on combined food subtests (out of 125), $n = 115$/fluid subtests (out of 50), $n = 107$ (excluding swallow items); CI, confidence interval; CPFQ, Queensland Cerebral Palsy Child Feeding Questionnaire; DDS, Dysphagia Disorders Survey; PSAS, Pre-Speech Assessment Scale; SOMA, Schedule for Oral Motor Assessment; t-cup, trainer-cup.

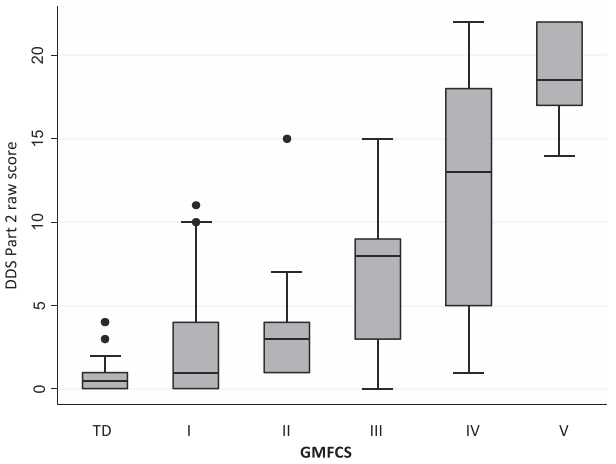

Fig. 1. Oropharyngeal dysphagia severity (Dysphagia Disorders Survey part 2) according to gross motor functional level (GMFCS). Key: linear regression showed significantly higher scores for all GMFCS levels when compared to the children with TD (GMFCS I $\beta = 1.6$, $p = 0.02$; II $\beta = 3.1$, $p < 0.01$; III $\beta = 6.0$, $p < 0.01$; IV $\beta = 11.3$, $p < 0.01$; V $\beta = 18.4$, $p < 0.01$). DDS, Dysphagia Disorders Survey; GMFCS, Gross Motor Function Classification System; TD, typically developing.

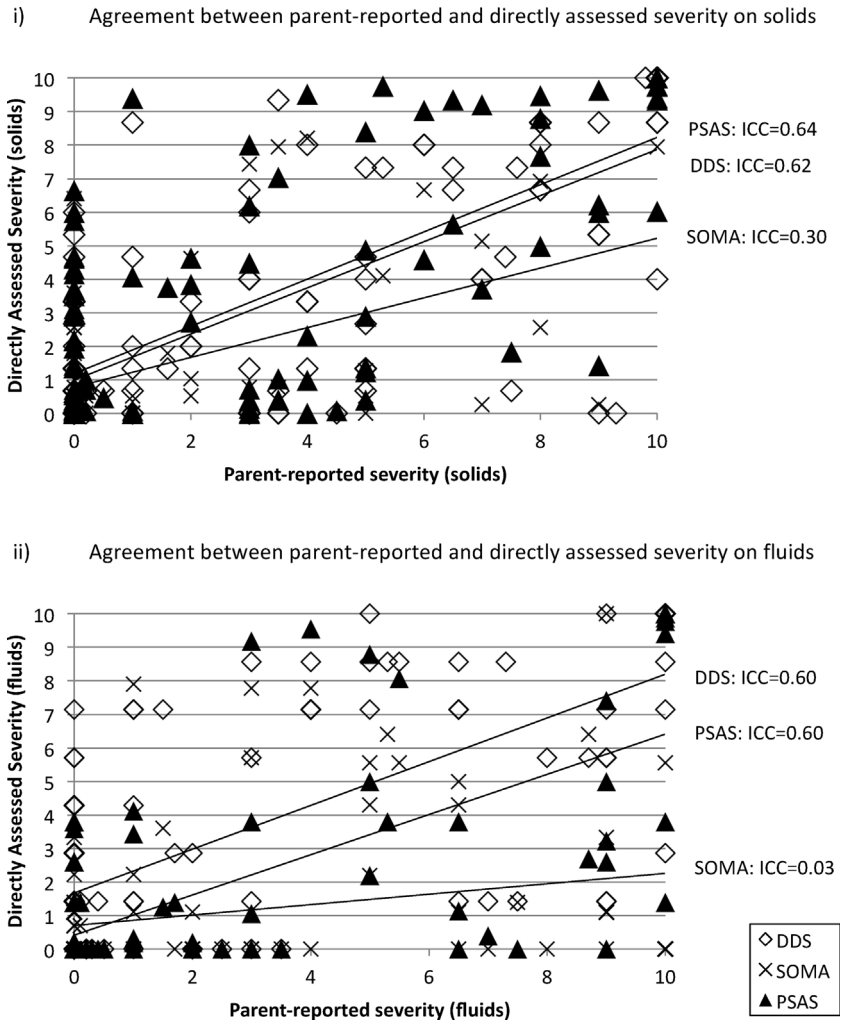

Fig. 2. Agreement between parent-reported and directly assessed oropharyngeal dysphagia severity in preschool children with cerebral palsy. Key: mean of differences for parent-reported OPD on solids and (i) DDS = 0.0 (SD = 2.9), (ii) SOMA = 2.4 (SD = 4.0), (iii) PSAS = 0.3 (SD = 3.6); mean of differences for parent-reported OPD on fluids and (i) DDS = −0.7 (SD = 2.9), (ii) SOMA = 1.5 (SD = 3.6), (iii) PSAS = 0.9 (SD = 3.2). DDS, Dysphagia Disorders Survey, ICC, Intra-class Correlation Coefficient; PSAS, Pre-Speech Assessment Scale; SOMA, Schedule for Oral Motor Assessment

5. Discussion

Almost all children with CP (over 90%) had directly assessed impairments to the oral phase of feeding, with the only children not classified as OPD belonging to GMFCS I. This finding was similar to that reported in children with CP aged 2–16 years by Kim and colleagues, although their sample was small ($n = 29$) (Kim et al., 2013). Generally, children did not have impaired saliva control in isolation of oral phase impairments in eating/drinking, although the reverse was true in half of the children. About 20% of these 'impairments' may be associated with typical development, as was found when the prevalence was calculated using the modified cut-points based on the typically developing reference group. This modified prevalence was equivalent to the proportion of children with difficulty eating or drinking reported by parents (about 80%), although the VAS was not restricted to oral phase impairments alone. There were more children with oral phase OPD with poorer GMFCS function, which was consistent with oral phase findings by Kim and colleagues (Kim et al., 2013). This was also consistent with our previous work (Benfer et al., 2013), and that by others (Calis et al., 2008; Erkin, Culha, Ozel, & Kirbiyik, 2010; Fung et al., 2002; Parkes, Hill, Platt, & Donnelly, 2010; Reilly et al., 1996; Santoro et al., 2012; Sullivan et al., 2000; Waterman, Koltai, Downey, & Cacace, 1992) looking more broadly at OPD.

The OPD severity of children with CP as a group was on average 3 out of 10 for solids and between 1 and 3 out of 10 for fluids. The DDS, PSAS and parent-reported average severity scores for solids were all equivalent, which suggests that they are all measuring a similar construct. The SOMA raw scores indicated the mildest OPD for both solids and fluids, which is likely because this measure is only detecting more clinically significant OPD, and missing milder cases. All children with CP had significantly higher scores on the DDS compared to children with TD, even children from GMFCS I. There was a stepwise

Table 3
Prevalence of specific oral phase impairments in young children with cerebral palsy, overall and according to gross motor function: solids.

Ingestion function	DDS, *n* (%)	SOMA, *n* (%)	PSAS, *n* (%)	Total impaired % (CI)	Sub-group: TD/GMFCS
Orienting to bolus	Orientation (non-chewable/ chewable) 26 (20.8)	[e]Head orientation to spoon (pr, r1): 15 (14.4)	[e]Visual recognition of spoon 25 (19.2)	35.9 (27.5–44.4)	TD: 0.0 I: 9.1* II: 6.7* III: 0.0 IV: 45.5* V: 95.7*
Stripping spoon	Reception non-chewable 34 (28.6)	[e]Lower lip draws inwards around spoon (pr, sd l1): 22 (20.6) [e]Upper lip removes food from spoon (pr, sd l2): 25 (23.4) [e]Graded jaw opening (pr, ss, sd j1): 42 (37.2)	[e]Lower lip draws inward around spoon: 18 (15.1) [e]Upper lip removes food from spoon: 21 (17.6)	52.3 (43.6–61.0)	TD: 2.5 I: 33.3* II: 66.7* III: 52.2* IV: 75.0* V: 78.3*
Biting	Reception chewable 67 (57.8)	[e]Controlled sustained bite (ck b5): 50 (46.3) Associated head movements to bite (ck j11): 42 (38.9) [e]Graded jaw opening (ck b8): 18 (16.7) [e]Lips close around stimulus during bite (ck l7): 42 (38.9) Mouths cracker only (ck b12): 1 (0.9)[b]	[e]Controlled sustained bite hard cookie: 43 (34.1) Associated movements with bite: 25 (19.7) [e]Graded jaw opening (24+): 71 (55.9)	70.0 (62.0–78.0)	TD: 12.5 I: 47.4* II: 66.7* III: 87.0* IV: 91.7* V: 100.0*
Saliva loss during eating	Containment[a]	Consistent/considerable drooling (ss, sd, ck d1): 25 (21.6)	NA	52.3 (43.6–61.0)[c]	TD: 17.5 I: 38.6* II: 26.7 III: 47.8* IV: 91.7* V: 87.0*
Food loss during eating	Containment 62 (51.2)	[e]Food loss none/trivial (ck fl1): 12 (11.2)	[e]Swallows with no food/ saliva loss: 51 (39.2)	54.6 (45.9–63.3)	TD: 5.0 I: 28.1* II: 60.0* III: 60.9* IV: 75.0* V: 100.0*
Cleaning behaviours	NA	[e]Lower/upper lip assists in cleaning (pr l3): 20 (19.2)	[e]Upper incisors to clean lower lip pom_pruicln: 47 (39.5) [e]Tongue is used to clean lips pom_prtcln: 83 (69.8)	70.0 (61.7–78.3)	TD: 35.0 I: 56.9* II: 53.3 III: 72.3* IV: 80.0* V: 100.0*
Oral transport: spoonable foods	Oral transport (non-chewable) 34 (28.6)	[e]Smooth rhythmic sequence (pr, ss sq1): 21 (19.1) [e]Sequence initiated within 2 seconds (ss i1): 5 (6.9) [e]Lower lip active during suck/chew/ munch (pr l11): 23 (22.1) Considerable/consistent tongue protrusion (pr t11): 25 (24.0) Protrusion beyond incisors (pr t12): 19 (18.3) [e]Internal jaw stabilisation (ss j2): 16 (22.3)	[e]No tongue protrusion (semi-solid swallowing subtest) (24 + m): 44 (33.9)	36.9 (28.5–45.3)	TD: 2.5 I: 14.0 II: 6.7 III: 39.1* IV: 58.3* V: 100.0*

Table 3 (*Continued*)

Ingestion function	DDS, *n* (%)	SOMA, *n* (%)	PSAS, *n* (%)	Total impaired % (CI)	Sub-group: TD/GMFCS
		External stabilisation required (ss j3): 16 (22.3) Associated jaw movements (ss j10): 16 (21.9)			
Oral transport: chewable foods	Oral transport (chewable) 44 (37.6)	[e]Smooth rhythmic sequence (sd sq1): 1 (7.7) [e]Sequence initiated within 2 seconds (ck i1): 6 (5.6) Lower lip behind upper teeth to suck (sd, ck l4): 17 (15.6) [e]Lower lip active during suck/chew/munch (sd l11): 1 (7.7) [e]Transient minimal tongue protrusion (sd, ck t10): 23 (20.9) Considerable/consistent tongue protrusion (ck t11): 23 (20.9) Protrusion beyond incisors (ck t12): 23 (20.9) Protrusion beyond lips (ck t13): 10 (9.1) [e]Internal jaw stabilisation established (ck j2): 22 (20.2) Variable stabilisation (ck j3): 25 (22.7) External stabilisation required (ck j4): 21 (19.1) Uses fingers to transfer food (ck j12): 22 (20.0)	[e]No tongue protrusion (24 + m): 53 (40.8)	55.0 (46.3–63.7)	TD: 12.5 I: 25.0 II: 33.3 III: 78.3* IV: 91.7* V: 100.0*
Chewing	Chewing 48 (41.0)	[e]Intermittent lip closure during munch/chew (ck l9): 14 (12.8) Vertical movements (ck j5): 2 (1.8) Wide vertical excursions (ck j8): 10 (9.1) [e]Small vertical excursions (ck j9): 10 (9.1)	Impairments to subtests[d]: Jaw chewing: 75 (60.5) Lips chewing: 53 (42.7) Tongue chewing: 65 (52.4)	65.4 (57.0–73.7)	TD: 22.5 I: 42.9* II: 60.0* III: 78.3* IV: 91.7* V: 100.0*

[a] Unable to separate drooling from food loss in DDS item therefore included only in food containment.
[b] Most children who are unable to bite cracker were not assessed on this texture.
[c] Totals include Thomas-Stonell and Greenberg Saliva Scale, proportion = 41.4%.
[d] Indicates delay to subtest greater than 1 month of age; SOMA items exclude those pertaining to the swallow (pharyngeal phase) including lip closure during swallow (ss) and gagging (ck).
[e] Item is framed as ability rather than impairment, therefore the proportion with impairment is reported.
* Significantly greater proportion impairment on logistic regression ($p < 0.05$) in GMFCS level relative to TD sample.

increase in the OPD severity (DDS score) as gross motor function declined, with this variable accounting for much of the variation in children's feeding scores. Children in GMFCS IV had the most variability between children in performance, with scores across almost the complete range of possible scores on the DDS (1–22).

The agreement between direct assessment and parent-reported oral phase impairments was only fair to moderate, although the statistics indicated this agreement was not simply by chance. The exception was the SOMA's drinking severity and detection of difficulty on cup, which had poor agreement. Even in the SOMA's original validation, the authors found poorer detection of impairments for the fluid subtests (Skuse, Stevenson, Reilly, & Mathisen, 1995). Interestingly, this agreement between impairment on the SOMA cup/trainer cup and a specific question related to parent reported 'difficulty on cup/trainer cup' was the highest agreement (80%). This may be due to under-detection by both the SOMA and parents for this particular item. Greater agreement was also found between 'difficulty moving food to back of mouth from spoon' (parent report) compared to this same task (oral transport of non-chewables) on the DDS (76%). Impairment to this task was only present in a small number of children (13–28%), and this impairment probably represents children with more overt difficulties, hence the better agreement. The direct measures all detected a similar proportion of children with difficulty biting/chewing (higher for the DDS and PSAS which looked at biting and chewing, whereas the SOMA was only biting). Parents only detected about half the directly detected impairment of this task, which suggests such an item should be worded more specifically if we want parents to identify equivalent impairments to the direct assessment.

Difficulty biting and absence of cleaning behaviours (of food from lips with upper incisors or tongue) were the most common specific oral phase impairments in our sample (both present in 70% of children with CP), followed closely by impaired chewing (65%). These three impairments were also commonly 'impaired' in the TD sample, so these figures could

Table 4
Prevalence of specific oral phase impairments in young children with cerebral palsy, overall and according to gross motor function: fluids.

	DDS, *n* (%)	SOMA, *n* (%)	PSAS, *n* (%)	Total impaired % (CI)	Sub-group: TD/GMFCS
Orienting to the bolus	Orientation (fluid): 17 (15.2)	[b]Anticipatory mouth opening (bottle r2): 5 (23.8)	NA	16.1 (9.2–23.0)	TD: 0.0
		No liquid enters the mouth (bottle r4): 1 (4.8)			I: 0.0
		[b]Accepts within 2 s (bottle, cup a2): 7 (9.9)			II: 14.3*
		Panic reactions when liquid presented (t-cup, cup sq2): 4 (4.2)			III: 5.6*
					IV: 36.4*
					V: 78.6*
Stripping teat (bottle), *n* = 21	NA	Upper lip seals firmly around teat (l3): 6 (28.6)	Bottle 'dysfunction' score: 8 (38.1)	47.6 (24.3–70.9)	TD: 0.0
		Intermittent/incomplete upper lip contact/seal (l5): 5 (23.5)			I: NA
		Intermittent/incomplete lower lip contact/seal (l6): 6 (28.6)			II: 16.7*
					III: NA
					IV: 83.3*
					V: 100.0*
Sipping from cup, *n* = 66	Reception (fluid)[a]: 42 (37.5)	NA	[b]Sucking pattern: 29 (22.3)	60.3 (51.5–69.2)	TD: 17.5
			External jaw stabilisation biting down on edge of cup: 8 (6.2)		I: 32.7
			[b]Internal jaw stabilisation: 55 (50.9)		II: 50.0*
			Tongue under cup: 4 (3.7)		III: 89.5*
					IV: 81.8*
					V: 100.0*
Liquid loss	Containment (fluid): 37 (33.0)	Profuse/marked liquid loss (>25%) (t-cup, cup ll): 21 (22.1)	Loses liquid bottle: 6 (4.6)	33.1 (24.9–41.3)	TD: 17.5
			Loses liquid cup (24+): 19 (14.6)		I: 19.3
					II: 40.0
					III: 21.7
					IV: 58.3*
					V: 60.9*
Oral transport	Oral transport 32 (28.6)	Small vertical movements (bottle, t-cup, cup j1): 12 (11.1)	Extension-retraction patterns (24+): 54 (41.5)	52.1 (43.0–61.1)	TD: 7.5
		[b]Smooth sequence (bottle sq1): 5 (23.8)			I: 23.6*
		Tongue thrusting (t-cup, cup t10): 7 (7.4)			II: 26.7*
		Asymmetry (t-cup, cup t11): 1 (1.1)			III: 77.8*
		Jaw clenching (cup j4): 1 (1.9)			IV: 90.1*
		Jaw alignment during drinking (t-cup j6): 7 (10.6)			V: 100.0*
		External stabilisation required (t-cup j10): 9 (13.6)			
		Internal jaw stabilisation (t-cup j12): 10 (15.2)			
		Uses gravity (t-cup s6): 17 (25.4)			
		Numerous attempts to initiate swallow (t-cup s7): 5 (7.5)			

[a] Includes sipping from any fluid utensil; SOMA items exclude those pertaining to the swallow (pharyngeal phase) including lip closure during swallow (bt), jaw alignment swallow (sw1), panic reaction after swallow (sw4), no swallow (sw5), choking (sq3), gagging (sw9).
[b] Item is framed as ability rather than impairment, therefore the proportion with impairment is reported.
* Significantly greater proportion impairment on logistic regression ($p < 0.05$) in GMFCS level relative to TD sample.

Table 5
Feeding frequency, duration and efficiency of preschool children with cerebral palsy, by gross motor function (GMFCS).

	Mean DDS score ±SD	Mean feeding frequency per day	*B* (*p* value)	Mean feeding duration per day	*B* (*p* value)	Feeding efficiency (kJ per minute)	*B* (*p* value)	Feeding efficiency (grams per minute)	*B* (*p* value)
GMFCS I, (*n* = 50)									
Actual	2.6 ± 3.0	7.1 ± 1.5	Ref.	118.2 ± 50.0	Ref.	38.8 ± 18.2	Ref.	8.4 ± 4.2	Ref.
Estimated	NA	NA	–	143.8 ± 72.1	Ref.	37.3 ± 24.8	Ref.	9.0 ± 5.4	Ref.
GMFCS II, (*n* = 13)									
Actual	4.1 ± 4.1	6.9 ± 1.6	−0.2 (0.74)	134.0 ± 36.7	15.8 (0.55)	31.6 ± 7.9	−7.2 (0.25)	7.2 ± 3.1	−1.2 (0.52)
Estimated	NA	NA	–	136.0 ± 37.5	−7.8 (0.73)	31.1 ± 7.7	−6.2 (0.42)	7.9 ± 3.5	−1.1 (0.62)
GMFCS III, (*n* = 19)									
Actual	7.1 ± 4.5	6.9 ± 1.8	−0.2 (0.74)	117.7 ± 43.8	−0.5 (0.98)	39.2 ± 19.6	0.4 (0.94)	9.1 ± 6.5	0.7 (0.64)
Estimated	NA	NA	–	136.1 ± 95.0	−7.7 (0.70)	37.0 ± 25.3	−0.3 (0.97)	9.9 ± 10.9	0.9 (0.65)
GMFCS IV[a], (*n* = 8)									
Actual	11.4 ± 7.5	6.8 ± 1.6	−0.3 (0.68)	92.8 ± 66.8	−25.4 (0.51)	41.5 ± 47.0	2.7 (0.74)	11.5 ± 15.6	3.1 (0.18)
Estimated	NA	NA	–	149.5 ± 73.5	5.8 (0.85)	41.0 ± 52.3	3.7 (0.71)	13.5 ± 17.5	4.5 (0.14)
GMFCS V[a], (*n* = 8)									
Actual	17.6 ± 2.3	7.0 ± 1.4	−0.1 (0.85)	132.8 ± 68.7	14.6 (0.58)	28.4 ± 17.1	−10.4 (0.17)	7.4 ± 3.7	−1.0 (0.66)
Estimated	NA	NA	–	158.4 ± 49.2	14.6 (0.58)	28.7 ± 17.3	−8.6 (0.34)	7.7 ± 3.7	−1.4 (0.61)
Partial tube, (*n* = 8)									
Actual	16.7 ± 6.3	6.1 ± 2.3	−1.0 (0.17)	105.0 ± NC	−13.2 (0.80)	13.8 ± 12.4	−25.0 (<0.01)	3.4 ± 4.2	−5.0 (0.03)
Estimated		NA		137.2 ± 126.1	−6.6 (0.84)	15.2 ± 12.7	−22.1 (0.02)	4.1 ± 4.7	−4.9 (0.08)

Key: *n* = 110 with completed food records, 4 were 100% tube fed, therefore excluded from this analysis (*n* = 106); actual time *n* = 37 (GMFCS I = 15, II = 5, III = 9, IV = 2, V = 5, tube = 1).

[a] Only completely orally fed children; β values show linear regression for mealtime outcomes and GMFCS; DDS, Dysphagia Disorders Survey; GMFCS, Gross Motor Function Classification System; kJ, kilojoules; NA, not applicable, as only actual values available; NC, confidence interval not calculable as *n* = 1; ref., reference group for analysis.

be inflated by limitations to oral tasks associated with typical development. Ability to do a sustained bite requires the child to have developed graded jaw movement, as well as adequate muscle strength to be able to break through the food without overflow movements (Morris, 2003). Impairments to these skills have been noted in the literature (Erkin et al., 2010). Other literature has suggested that tasks performed on the midline (such as biting and spoon feeding) were better than those requiring multiple planes of movement (such as chewing and cup drinking) (Gisel et al., 2000; Yilmaz et al., 2004). Oral transport of purees and orienting were least frequently impaired, consistent with the clinical picture that these skills are likely to be associated with more severe OPD.

Difficulty actively sipping from a cup (with a stable jaw) was the most frequently observed impairment with fluids (in 60% of children), followed by difficulty in oral transport of fluids (in half) and stripping a bottle teat (in half of those assessed on a bottle). Liquid loss was quite common even in the children with TD (20%), but was less common in GMFCS IV–V than other impairments, perhaps due to parents positioning their child (reclined or with head back) and/or limiting bolus delivery (smaller and single sips only, and modified utensils) in this group. Generally there were more children with CP who had specific oral phase impairments compared to children with TD, with the exception of oral transport of solids (only significant for GMFCS III–V) and liquid loss (significant for GMFCS IV–V). Looking at the raw data, it appears that fewer children from GMFCS III have liquid loss; however, it is important to note that the DDS does not account for children's use of different fluid utensils. When we look at the utensil use of children, we see that most children in level III are using a trainer cup or bottle, which limits the flow rate, thus making the fluids easier to manage orally with less liquid loss. The number of children not using a cup was significantly higher for children with CP compared to the children with TD, which may obscure some of the differences in skills between these groups.

Oral phase impairments are known to reduce the efficiency of bolus processing (Gisel, 1988), which may lead to prolonged mealtimes (Dahl, Thommessen, Rasmussen, & Selberg, 1996; Sullivan et al., 2000; Waterman et al., 1992; Wilson & Hustad, 2009), with mealtime duration suggested to be a reliable measure of OPD severity (Sullivan et al., 2000). Our study findings showed no difference in the number of meals, average daily feeding duration or feeding efficiency (kJ per minute or grams per minute) based on gross motor level or OPD severity. The exception was the reduced feeding efficiency (kJ and grams) of children with partial tube feedings, all of whom were from GMFCS IV and V. There was a lot of variability of the duration/frequency/efficiency variables within GMFCS levels (and OPD scores), suggesting that parents of a child with significant oral phase impairments may have had modifications to the texture or energy density of their diet, and consequently be performing well for their level. As children's mealtime efficiency (different from the 'feeding efficiency' of an individual bolus, as reported by Gisel (1988)) also was not significantly different with more severe OPD, we can propose that the modifications made to the textures in children's diets (Benfer, 2014 [unpublished work]) on the whole may be adequate to maintain their efficiency of intake. Our previous

work also found a reduced energy intake was associated with children with lower gross motor function (Benfer, 2014 [unpublished work]), so interventions to support overall energy intake in children with poorer gross motor function may be indicated.

This study represents the first to our knowledge to document the patterns of oral phase impairments using standardised OPD measures, according to GMFCS, across the full spectrum of gross motor function. While the use of standardised OPD measures is a step forward in understanding the clinical presentation of children with CP, there were a number of limitations surrounding these measures. While the measures were focused predominately on the oral phase, a couple of the items in the DDS and SOMA pertained to the pharyngeal phase, which may influence the overall oral phase prevalence estimates. In addition, due to the lack of a gold standard measure, we reported a combined total estimate based on a positive OPD classification on one or more of the DDS, SOMA or PSAS which may inflate the estimate. Some measures had multiple items pertaining to a single oral task, which may inflate the prevalence of that particular task relative to other oral tasks, so this needs to be considered when interpreting the findings. The use of raw scores as measures of OPD severity have not been validated for this purpose, as a stepwise change in scores may not represent a linear change in severity, particularly for the SOMA whose scoring was structured to be a discriminative measure rather than evaluative. Finally, using the three day weighed food record, an already high-burden data collection method, to collect mealtime duration, meant that there were moderate levels of missing data across the records. To account for this, we reported on both actual and estimated mealtime durations, which did not differ significantly.

There are implications for clinicians and researchers with regards to therapy planning, screening (particularly using parent report) and nutritional management. The findings from this study will provide useful information to assist in planning therapy interventions, particularly specific oral sensorimotor approaches. Children with better gross motor function tended to have isolated oral phase impairments, whereas those with poorer gross motor function had more systematic impairments involving multiple oromotor subsystems. This data will also provide a foundation for future research on feeding therapies, in targeting specific approaches to specific CP sub-groups. Certain 'impairments' were present in a large number of children with TD, which should be considered in clinical management of children with CP, particularly those with mild impairments. These 'impairments' need to be examined more closely to understand if those observed in typical development can be differentiated from those representing delayed or disordered patterns, with regards to quality of the movement or frequency.

Parent report has been used extensively in research on OPD to date, although there is a limited understanding of the accuracy of this as a proxy for a direct objective assessment. This study suggested that parents were in agreement with direct assessment only about half the time, although they were not systematically under-reporting. More specifically worded questions and those asking about more overt OPD tended to have better agreement with direct assessment.

We found that children who were partially tube fed had significantly lower feeding efficiency, so this could be a useful early indicator of children needing supplementation to their nutrition (through increasing energy density of foods/fluids, or tube feeds). Our findings suggested that the child's OPD severity or gross motor level was not significantly influencing the efficiency of their mealtime, although an association may be obscured by parents who are already modifying textures or energy density of their child's food to account for their OPD. Previous findings (Benfer, 2014 [unpublished work]) suggest a lower energy intake of children with lower gross motor function, thus parent education around increasing energy density of easier to manage textures, and an increased frequency of meals for these children may be important to improve their nutritional outcomes.

6. Conclusions

Oral phase OPD was common in preschool children with CP, present in 93.8% of children when directly assessed (78.5% with modified cut-points), and 79.2% based on parent report. The agreement between direct assessment and parent reported oral phase OPD was only fair, although parents did not consistently under-report. More specifically worded questions and those asking about more overt OPD tended to have better agreement with direct assessment, which should be considered when implementing parent-reported screening. OPD severity and GMFCS were not related to mealtime frequency, duration or efficiency, although children on partial tube feeds had significantly reduced mealtime efficiency. These findings highlight the importance of considering feeding efficiency as an early marker for children needing nutritional supplementation or modifications to their diet.

Competing interests

The authors declare they have no competing interests.

Funding

This project was supported by the National Health and Medical Research Council Postgraduate Medical and Dental Scholarship (1018264 – KB), Career Development Fellowship (APP1037220 – RB) and Project Grants (569605 and 465128).

Acknowledgements

We would like to thank Physiotherapists Rachel Jordan (BPT) and Chris Finn (BPT) for data collection and gross motor ratings and Dietitians Stina Oftedal (B.Hlth.Sc (Hons) Nutr & Diet) and Camilla Davenport (B.Hlth.Sc (Hons) Nutr & Diet) for data collection of feeding videos.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at http://dx.doi.org/10.1016/j.ridd.2014.08.029.

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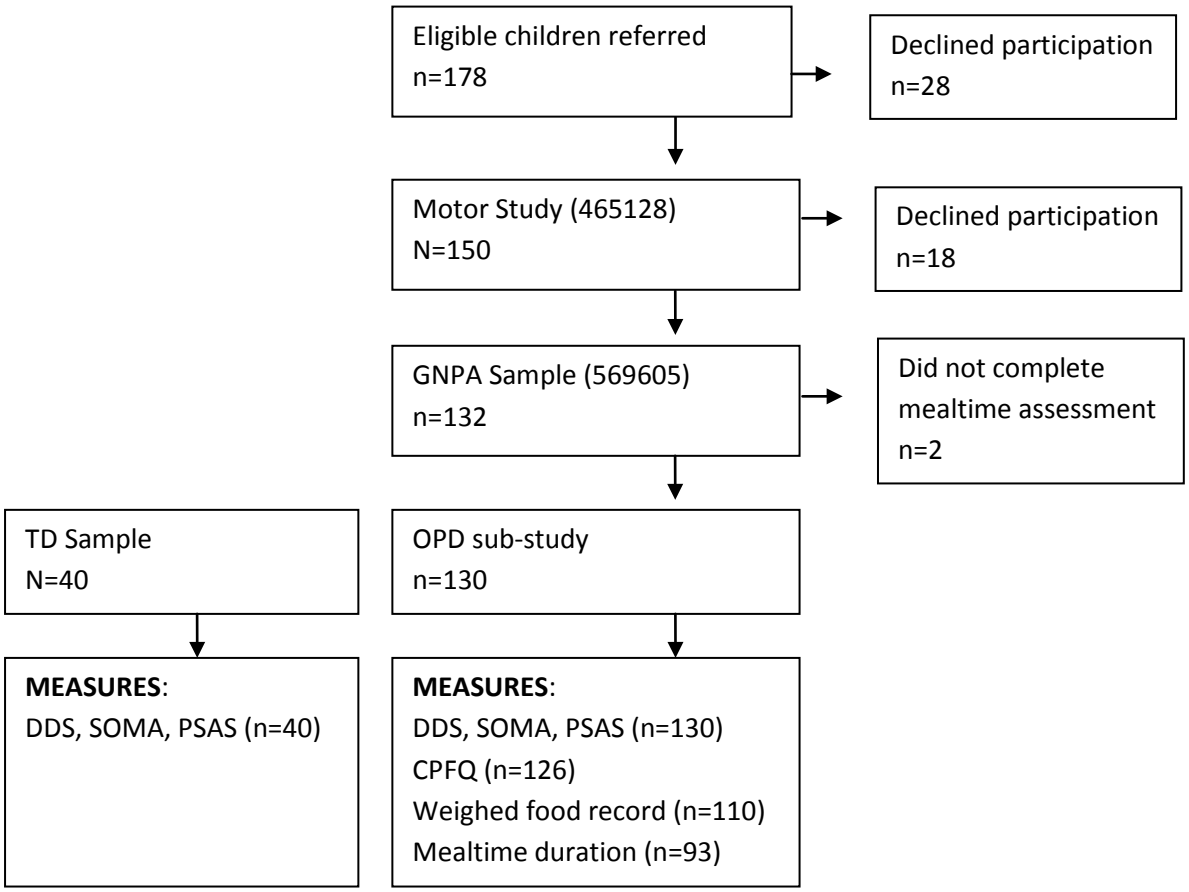
Supporting Information 1: Participant numbers for oropharyngeal dysphagia study



Key: CPFQ Queensland Cerebral Palsy Child Feeding Questionnaire ; DDS Dysphagia Disorders Survey; GNPA Growth Nutrition and Physical Activity; OPD Oropharyngeal Dysphagia; PSAS Pre Speech Assessment Scale; SOMA Schedule for Oral Motor Assessment; TD Typically Developing

Supporting Information 2: Dysphagia Disorders Survey: Oral Phase Items according to Gross Motor Function

	TD	CP: GMFCS				
		I	II	III	IV	V
Non-chewable						
Orientation (%±CI)	0.0	0.0	6.7±10*	0.0	36.4± 10*	95.6±10 *
Reception (%±CI)	0.0	7.3±7.6*	20.0±21.4*	22.7±18.3*	54.5±31.4*	100.0±0.0*
Containment (%±CI)	0.0	12.7±9.7*	6.7±13.3*	36.4±21.0*	54.5±31.4*	100.0±0.0*
Oral Transport (%±CI)	0.0	9.1±7.8*	6.7±13.3*	22.7±18.3*	63.6±30.4*	100.0±0.0*
Chewable						
Orientation (%±CI)	0.0	0.0	6.7±13.3*	4.4±8.7*	45.5±31.4*	81.3±20.1*
Reception (%±CI)	7.5±8.4	35.7±12.5*	53.3±26.6*	69.6±20.0*	81.8±24.4*	87.5±17.0*
Containment (%±CI)	5.0±7.0	23.2±11.4*	53.3±26.6*	52.2±21.4*	63.6±30.4*	68.8±24.0*
Oral Transport (%±CI)	0.0	10.7±8.4*	20.0±21.4*	60.9±20.8*	63.6±30.4*	87.5±17.0*
Chewing (%±CI)	0.0	14.3±9.4*	40.0±13.1*	52.2±10.7*	72.7±28.1*	87.5±17.0*
Fluids						
Orientation (%±CI)	0.0	0.0	6.7±13.2*	4.8±9.4*	36.4±30.4*	73.3±23.6*
Reception (%±CI)	5.0±6.9	14.5±9.4	26.7±23.6*	38.1±21.6*	72.7±28.0*	93.3±13.3*
Containment (%±CI)	17.5±12.0	14.5±9.4	40.0±26.2*	14.3±15.6	63.6±30.4*	86.7±18.2*
Oral Transport (%±CI)	0.0	9.1±3.9*	6.7±13.2*	28.6±20.2*	72.7±28.0*	86.7±18.2*

*Significantly different from TD on logistic regression (binary outcomes) or linear regression (continuous outcomes); CI Confidence Interval; CP Cerebral Palsy; GMFCS Gross Motor Function Classification System; TD Typically Developing

Summary of Chapter 7

This chapter provided an understanding of the patterns of oral phase impairments common in preschool children with CP, and how this varies by gross motor function.

i. Almost all children with CP had oral phase impairments during eating, drinking or controlling saliva (93.8%, using modified cut-points 78.5%). Children generally did not have impaired saliva control in isolation of other oral phase impairments.
ii. Impairments were present in children from all GMFCS levels, including those with ambulatory CP. GMFCS was strongly associated with impairments of the oral phase, with all children from GMFCS II-V having 1 or more oral phase impairments.
iii. The most commonly observed oral phase impairments for children with CP were impaired biting (70%), absence of cleaning behaviours (eg, scraping with incisors or licking with tongue, 70%), and impaired chewing (65%). These were also frequently 'impaired' in children with TD, although less than the CP sample (13%, 35%, 23%, respectively).
iv. Children from all levels of gross motor function (GMFCS I-V) had significantly higher scores on the DDS compared to children with TD, indicating impairment across a greater number of ingestion functions.
v. Agreement between parent-report and direct assessment was only moderate (39% to 63%), although parents were not consistently overreporting or underreporting (for severity of oral phase). This appeared to be influenced by whether parents thought their child was unsafe on the texture rather than impaired.
vi. There were more children with CP who used infant bottles and trainer cups compared to children with TD. The proportion of children who did not use an open cup increased with increasing GMFCS level.
vii. There were no differences in the daily average number of meals, average daily feeding duration, or feeding efficiency (grams or kilojoules per minute) based on gross motor function or OPD severity. There was a lot of variability of these factors within GMFCS levels, suggesting some parents may be already modifying their child's diet (texture or energy density), which has influenced these findings.

Impairments associated with eating and drinking are commonly described according to the oral phase and pharyngeal phase of the swallow. As such, this detailed description of the impairments of the oral phase is complemented by the following chapter, which describes the clinical signs associated with pharyngeal phase dysphagia.

Chapter 8: Clinical Signs Suggestive of Pharyngeal Dysphagia in Preschool Children with Cerebral Palsy

Introduction to Chapter 8

In keeping with the detailed examination of the oral phase impairments, this chapter examines the clinical signs suggestive of pharyngeal phase impairment in preschool children with CP. Consistent with the previous chapter, this analysis is performed based on each level of gross motor function (on the GMFCS). This is by way of the article (in press) "Clinical Signs Suggestive of Pharyngeal Dyspagia in Preschool Children with Cerebral Palsy". This article reports on 16 clinical signs suggestive of pharyngeal phase OPD, including discriminative validity (by comparison to a sample of children with TD), reproducibility of signs (interrater and intrarater) and prevalence data. Videofluoroscopic swallow study data were available for only n=9 children (within 12 months of the child's clinical assessment) which was judged to be inadequate to provide concurrent validity for the clinical signs in the publication. This data has been included at the end of the chapter for completeness (Table 6).

Paper 7: Clinical Signs Suggestive of Pharyngeal Dysphagia in Preschool Children with Cerebral Palsy

This article was published in Research in Developmental Disabilities (journal impact factor 2.735). Reprinted from Research in Developmental Disabilities, Vol 38, Benfer, K. A., Weir, K. A., Bell, K. L., Ware, R. S., Davies, P. S. W., & Boyd, R. N., Clinical signs suggestive of pharyngeal dysphagia in preschool children with cerebral palsy, pages no. 192-201, Copyright 2015, with permission from Elsevier.

> Benfer K.A., Weir K.A., Bell, K.L., Davies, P.S.W., Ware, R.S., Boyd R.N. Clinical signs suggestive of pharyngeal dysphagia in preschool children with cerebral palsy. *Res. Dev. Disabil.* 2015;38:192-201.

This paper was also presented at the 6th Biennial Conference of the Australasian Academy of Cerebral Palsy and Developmental Medicine, 30 May-1 June 2012, Brisbane, Australia (as a free paper); and the 65th Annual Meeting of the American Academy of Cerebral Palsy and Developmental Medicine, 12-15 October 2011, Las Vegas, United States (as a poster).

Benfer K.A., Weir K.A., Bell, K.L., Davies, P.S.W., Ware, R.S., Boyd R.N. Reported and observed clinical signs of oropharyngeal aspiration in young children with cerebral palsy. *Dev. Med. Child Neurol.* 2012;54(Supp 5):21. (Abstract)

Benfer, K.A., Weir, K.A., Bell, K.L., Robinson, P.M., Davies, P.S.W., Ware, R., Boyd, R.N. Reported and observed clinical signs of oropharyngeal aspiration in young children with cerebral palsy. *Dev. Med. Child Neurol.* 2011;53(Supp 3):23. (Abstract)

Research in Developmental Disabilities 38 (2015) 192–201

Contents lists available at ScienceDirect

Research in Developmental Disabilities

Clinical signs suggestive of pharyngeal dysphagia in preschool children with cerebral palsy☆

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ARTICLE INFO

Article history:
Received 14 October 2014
Received in revised form 10 December 2014
Accepted 11 December 2014
Available online

Keywords:
Orpharyngeal dysphagia
Deglutition disorders
Pharyngeal phase impairment
Oropharyngeal aspiration
Cerebral palsy
Children

ABSTRACT

This study aimed to determine the discriminative validity, reproducibility, and prevalence of clinical signs suggestive of pharyngeal dysphagia according to gross motor function in children with cerebral palsy (CP). It was a cross-sectional population-based study of 130 children diagnosed with CP at 18–36 months (mean = 27.4, 81 males) and 40 children with typical development (TD, mean = 26.2, 18 males). Sixteen signs suggestive of pharyngeal phase impairment were directly observed in a videoed mealtime by a speech pathologist, and reported by parents on a questionnaire. Gross motor function was classified using the Gross Motor Function Classification System. The study found that 67.7% of children had clinical signs, and this increased with poorer gross motor function (OR = 1.7, $p < 0.01$). Parents reported clinical signs in 46.2% of children, with 60% agreement with direct clinical mealtime assessment (kappa = 0.2, $p < 0.01$). The most common signs on direct assessment were coughing (44.7%), multiple swallows (25.2%), gurgly voice (20.3%), wet breathing (18.7%) and gagging (11.4%). 37.5% of children with TD had clinical signs, mostly observed on fluids. Dysphagia cut-points were modified to exclude a single cough on fluids, with a modified prevalence estimate proposed as 50.8%. Clinical signs suggestive of pharyngeal dysphagia are common in children with CP, even those with ambulatory CP. Parent-report on 16 specific signs remains a feasible screening method. While coughing was consistently identified by clinicians, it may not reflect children's regular performance, and was not sufficiently discriminative in children aged 18–36 months.

☆ ANZTR Trial Registration Number: ACTRN12611000616976.

Abbreviations: CP, cerebral palsy; CPFQ, Queensland Cerebral Palsy Feeding Questionnaire; GMFCS, gross motor function classification system; GNPA, Growth, Nutrition, and Physical Activity (study); NHMRC, National Health and Medical Research Council (Australia); OPD, oropharyngeal dysphagia; SD, standard deviation; TD, typical development; VFSS, Videofluoroscopic Swallow Study.

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http://dx.doi.org/10.1016/j.ridd.2014.12.021
0891-4222/

1. Introduction

Oropharyngeal aspiration (food or fluid entering the trachea below the vocal folds) (Brockett, 2006) is a commonly cited risk factor for recurrent pneumonia (Vaughan & Katkin, 2002) occurring frequently in children with non-ambulatory cerebral palsy (CP) and oropharyngeal dysphagia (OPD) (Mirrett, Riski, Glascott, & Johnson, 1994). In addition to causing pneumonia, chronic aspiration may lead to interstitial lung disease, pulmonary fibrosis and bronchiectasis (Lefton-Greif & McGrath-Morrow, 2007; Vaughan & Katkin, 2002). The nature of the aspirate, amount and frequency of aspiration all influence the consequent health outcomes, although the progression of respiratory sequelae and prognosis in children with CP are poorly understood (Cass, Wallis, Ryan, Reilly, & McHugh, 2005; Lefton-Greif & McGrath-Morrow, 2007). Factors related to respiratory status are of utmost importance, as respiratory-related factors are a leading cause of premature mortality in individuals with CP (Blair, Watson, Badawi, & Stanley, 2001). CP is a motor disability arising from a non-progressive neurological lesion, impacting on the strength and coordination of motor control (Smithers-Sheedy et al., 2013). As such, neurologically mediated mechanisms can compromise the sensorimotor tasks of eating and drinking; with impairments (OPD) occurring at any of the four phases of swallowing, including the oral-preparatory, oral-propulsive, pharyngeal and oesophageal phases (Matsuo & Palmer, 2008).

The pharyngeal phase involves a complex set of sensory and motor responses as food or fluid pass through the pharynx (Matsuo & Palmer, 2008). The pharynx is a shared anatomical juncture, involved in the functions of swallowing and respiration; hence, airway protection to prevent aspiration before, during and after bolus passage through the pharynx is critical for respiratory health. Airway protection is achieved by closure of the true and false vocal folds, the epiglottis inverting in response to the hyo-laryngeal excursion during the swallow, and finally deglutitive expiratory airflow (glottal release) (Lefton-Greif & McGrath-Morrow, 2007). Pharyngeal phase impairments in children with neurological conditions include inadequate airway protection during the swallow, incomplete laryngeal clearance following the initial swallow efforts, and/or decreased strength of pharyngeal contraction resulting in persistent residue in the hypopharynx post-swallow (Morton, Minford, Ellis, & Pinnington, 2002). Problems with the volitional oral motor movements of the oral-preparatory and propulsive phases may also affect bolus transit and thus compromise airway protection.

In addition to the specific neurophysiological limitations to the oropharyngeal mechanism, OPD in children with CP is also associated with their gross motor function (Benfer et al., 2013; Calis et al., 2008; Fung et al., 2002; Parkes, Hill, Plat, & Donnelly, 2010; Reilly, Skuse, & Poblete, 1996; Sullivan et al., 2000; Waterman, Koltai, Downey, & Cacace, 1992). An unstable pelvis and trunk can result in poor head and neck positioning, reducing the ability for controlled oropharyngeal movements (Bosma, 1992; Langley & Thomas, 1991). Children with CP may use disordered patterns of movement to create a base of stability, such as scapular retraction, which can influence the position of the oropharyngeal structures and restrict their mobility (Arvedson, Brodsky, & Reigstad, 2002). Poor head position has also been related to compromised airway protection by opening the airway, and the influence of gravity on flow rate of foods/fluids swallowed (Arvedson et al., 2002; Ekberg, 1986; Lanert & Ekberg, 1995).

The safety of the swallow is initially screened for clinically, including a comprehensive evaluation of the mealtime, and observation of clinical signs suggestive of pharyngeal phase impairment. A number of clinical signs have been used to indicate aspiration, with varying levels of sensitivity/specificity when compared to instrumental assessment, depending on the texture being assessed (Arvedson, Rogers, Buck, Smart, & Msall, 1994; DeMatteo, Matovich, & Hjartarson, 2005; Rogers, Arvedson, Buck, Smart, & Msall, 1994; Warms & Richards, 2000; Weir, McMahon, Barry, Masters, & Chang, 2009). When food or fluid reaches the vocal folds, a protective cough may be triggered, although children with CP are at high risk of 'silent aspiration' (no coughing when foods/fluids are aspirated), reported in between 82% (Weir, McMahon, Taylor, & Chang, 2011) and 94% (Arvedson et al., 1994) of cases of aspiration. It is therefore important in this population to observe other signs of aspiration such as wet/gurgly respiration or phonation, and fremitus (rattly chest). A child with clinical indications of aspiration may have this confirmed through evaluation with videofluoroscopic swallow study (VFSS). While widely considered the gold standard for detecting aspiration, VFSS tends to be restricted to tertiary hospitals (requiring trained personnel) and children are exposed to radiation during the procedure. Thus referral rates have remained relatively low, depending on the geographical region (Clancy & Hustad, 2011; DeMatteo et al., 2005; Waterman et al., 1992).

A number of studies have explored the patterns of pharyngeal phase impairments in CP, using clinical (Arvedson et al., 1994; Calis et al., 2008; Dahl, Thommessen, Rasmussen, & Selberg, 1996; Del Giudice et al., 1999; Erkin, Culha, Ozel, & Kirbiyik, 2010; Fung et al., 2002; Gerek & Ciyiltepe, 2005; Reilly & Skuse, 1992; Reilly et al., 1996; Rogers et al., 1994; Santoro et al., 2012; Sullivan et al., 2000; Wilson & Hustad, 2009; Yilmaz, Basar, & Gisel, 2004) and instrumental assessments (Arvedson et al., 1994; Field, Garland, & Williams, 2003; Gisel, Applegate-Ferrante, Bensen, & Bosma, 1995; Griggs, Jones, & Lee, 1989; Helfrich-Miller, Rector, & Straka, 1986; Morton et al., 2002; Rogers et al., 1994; Waterman et al., 1992; Weir et al., 2007, 2011; Wright, Wright, & Carson, 1996), but estimates of specific clinical signs of pharyngeal phase impairment have varied significantly. Many of the studies identified clinical signs through parent-report and only recruited children with moderate-severe CP or those with OPD. Further, children were either school-aged or recruitment spanned a broad age range (from infancy to adolescence). Coughing and/or choking (17–100%) (Del Giudice et al., 1999; Gerek & Ciyiltepe, 2005), gagging (14–69%) (Rogers et al., 1994) (Wilson & Hustad, 2009), and regurgitation (2.5–45%) (Erkin et al., 2010) (Reilly et al., 1996) were most frequently reported. There is generally consensus from instrumental assessment that thin fluids are the most likely food/fluid consistency to be aspirated in children with CP (Arvedson et al., 1994; Gisel et al., 1995; Morton et al., 2002; Rogers et al., 1994; Weir et al., 2007, 2011).

The aforementioned studies provide a greater understanding of pharyngeal phase impairments in older children with moderate-severe CP, but there are no studies to our knowledge exploring this in a population-based sample of preschool-aged children with CP. The current study aimed to determine the discriminative validity, reproducibility and prevalence of clinical signs suggestive of pharyngeal dysphagia according to gross motor function in children with CP. In order to improve earlier screening of potential pharyngeal dysphagia in children with CP aged 18–36, we need to consider which signs are associated with typical development at this age, and therefore may not be robust indicators of impairment. Furthermore, signs which can not be reliably observed between clinicians or on multiple ratings by the same clinician may not be suitable to use in screening. It was hypothesised that clinical signs would be prevalent in more than half of the children with CP, across all gross motor levels (GMFCS I–V), and the proportion with signs increase with poorer gross motor function (GMFCS IV and V).

2. Materials and methods

This is a cross-sectional population-based study of preschool-aged children with CP, conducted in Queensland, Australia between April 2009 and March 2013. It is part of two concurrent longitudinal studies, the Queensland CP Child: Motor Function and Brain Development study (National Health and Medical Research Council, NHMRC 465128) (Boyd et al., 2013) and the Queensland CP Child: Growth, Nutrition and Physical Activity study (GNPA, NHMRC 569605) (Bell et al., 2010; Benfer et al., 2012). Ethics approvals have been reported in study protocol papers (Bell et al., 2010; Benfer et al., 2012; Boyd et al., 2013). All families gave written informed consent to participate.

2.1. Participants

Three samples were recruited for this study, as shown conceptually in Fig. 1. All children with a confirmed diagnosis of CP, aged 18–36 months corrected age, and born in Queensland between 2006 and 2009 were invited to participate in the GNPA sample (Bell et al., 2010). Children with neurodegenerative conditions were excluded.

Forty children with typical development (TD) were recruited through convenience sampling. Children were term births (>37 weeks), did not have a diagnosis requiring neonatal admission or ongoing medical/allied health treatment, and were not on regular medication. Recruitment was stratified by age (18–24 and 30–36 months).

Forty children (eight/GMFCS level) were selected randomly by an independent researcher for analysis of intra-rater and inter-rater reproducibility of clinical signs. Thirty of these children were from the GNPA sample, with an additional ten children recruited (included for pragmatic reasons, forming part of another study). These children also had a confirmed diagnosis of CP, aged 18–36 months corrected age, and those who were non-oral due to tube feeding were excluded.

2.2. Procedures

Children attended the hospital or were seen at home for direct mealtime assessment, according to the snack protocol (Benfer et al., 2012). Four signs (gurgly voice, wet breathing, fremitus, cough) were rated live pre- and post-mealtime by a

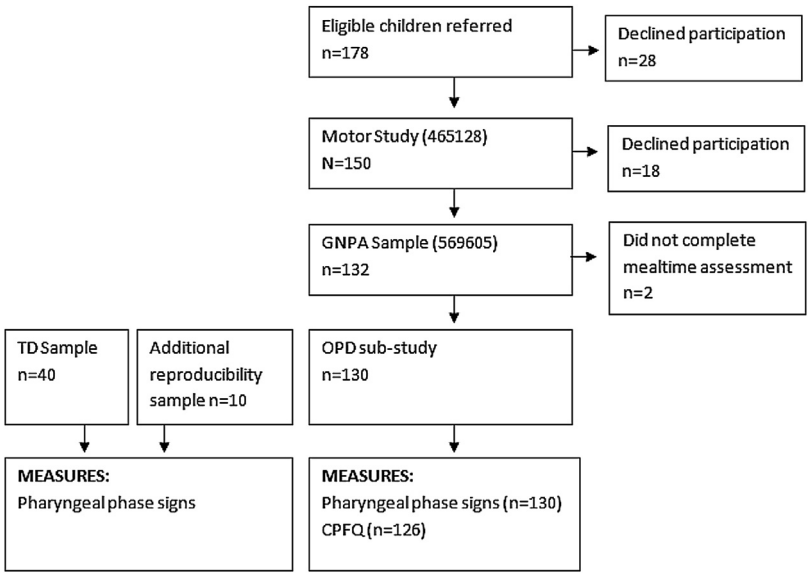

Fig. 1. Participant numbers for oropharyngeal dysphagia study: Growth Nutrition and Physical Activity, Reproducibility and Typically Developing Samples. Key: CPFQ Queensland Cerebral Palsy Child Feeding Questionnaire; GNPA, Growth, Nutrition and Physical Activity; OPD, oropharyngeal dysphagia; TD, typically developing.

researcher, and mealtimes were videoed for rating by a speech pathologist. Three standardised presentations of four textures (puree, semi-solid, chewable and fluid) were presented by the caregiver using the child's regular utensils before allowing completion of the snack. Children with CP also had gross motor assessment conducted by two physiotherapists.

2.3. Measures

2.3.1. Clinical signs suggestive of pharyngeal phase impairment

A determination of pharyngeal phase OPD was noted if children demonstrated any one of 16 signs, selected from the literature (DeMatteo et al., 2005; Lefton-Greif & McGrath-Morrow, 2007) and research conducted by our co-author (Weir et al., 2009). These signs included gagging, coughing, choking, vomiting, throat clearing, multiple swallows, wheezing, stridor, rapid or laboured breathing, wet breathing, gurgly voice, rattly chest, snuffly nose, eye tearing, or circumoral cyanosis/duskiness, noted for each food/fluid texture. Wet voice (sensitivity 0.67, specificity 0.92), wet breathing (sensitivity 0.33, specificity 0.83) and cough (sensitivity 0.67, specificity 0.53) were considered good clinical markers of oropharyngeal aspiration (compared to VFSS) on thin fluids, but not for pureed textures (Weir et al., 2009).

2.3.2. Parent-reported clinical signs suggestive of pharyngeal phase impairment

The same clinical signs were documented by parents on the Queensland Cerebral Palsy Child Feeding Questionnaire (CPFQ) according to written descriptions of terms (Appendix A, adapted from the Children's Feeding Skills Questionnaire, Royal Children's Hospital, Brisbane). Presence or absence of specific signs was prospectively recorded for each texture across a number of mealtimes.

2.3.3. Gross motor function

Children's gross motor function was classified according to the five levels of the GMFCS (Palisano et al., 1997) on the <2 year and 2–4 year age bands. Motor type (spastic, dyskinetic and hypotonic) and distribution (unilateral, bilateral, and number of limbs) were also classified according to the Surveillance of CP in Europe (Cans, 2000; Sanger, Delgado, Gaebler-Spira, Hallett, & Mink, 2003).

3. Calculations

All data analyses were performed using Stata 10.0 (Statcorp 2007), with significance set at $p < 0.05$. Demographic data were presented with descriptive statistics. Discriminative validity was determined through logistic regression for each sign compared to the TD sample. In order to account for clinical signs associated with typical development, signs observed in more than 25% of children with TD were excluded from the modified prevalence estimate. Inter- and intra-rater reproducibility were assessed using Cohen's Kappa and percentage agreement (overall and by specific sign). The prevalence of clinical signs suggestive of pharyngeal phase impairment (overall and of specific signs; based on direct-assessment and parent-report) were presented according to GMFCS. The agreement between direct-assessment and parent-report for each sign was reported with percentage agreement.

4. Results

There were 178 eligible children referred, of which 132 families consented to participate in the GNPA study, with 130 children completing the mealtime assessment (Fig. 1). Seven children defaulted to 'impaired' for overall fields in the direct-assessment (pharyngeal phase overall, and each texture overall) as they were unsafe to have all foods orally (indicated prior to research participation, by their primary medical team). There were 21 males (52.5%) in the sample of children with typical development, with a mean age of 27.4 months (SD = 6.0). The sample characteristics of the GNPA sample are shown in Table 1. The GNPA sample has been shown previously to be representative of the population of children with CP (Benfer et al., 2013).

4.1. Validation of clinical signs suggestive of pharyngeal phase impairment

The prevalence of clinical signs in children with TD was 37.5%, with 14/15 children with signs observed during ingestion of thin fluids. Ten of the children with clinical signs were aged 18–24 months and five were 30–36 months. Two different signs were observed in the children with TD; coughing ($n = 15$, 37.5%) and wet breathing ($n = 3$, 7.5%). Three children with TD had signs across multiple textures, (one on thin fluid and pureed food; and two on thin fluid, pureed and chewable foods) and a further two children had multiple coughs on a single texture. There were significantly more signs observed in children from GMFCS IV and V compared to the TD sample ($\beta = 0.9$, $p = 0.03$, $\beta = 3.6$, $p < 0.01$, respectively).

4.2. Reproducibility of clinical signs suggestive of pharyngeal phase impairment

Results of the reproducibility study are presented in Table 2. The inter- and intra-rater reproducibility of clinical signs were strong overall with >90% agreement between raters, and >95% for a single rater. Agreement was marginally better for intra-rater compared to inter-rater for each of the signs. The signs with the strongest agreement between raters were gag,

Table 1
Characteristics of participants of the Growth, Nutrition and Physical Activity Study.

	GNPA participants ($n = 130$)
Gender, males (n, %)	81 (62.3)
Age (mean, SD)	27.2 (5.4)
GMFCS level (n, %)	
I	57 (44.2)
II	15 (11.6)
III	23 (17.8)
IV	12 (9.3)
V	23 (17.7)
Primary motor type (n, %)	
Unilateral spasticity	41 (31.5)
Bilateral spasticity	72 (55.4)
Dystonia	2 (1.5)
Ataxia	2 (1.5)
Hypotonia	9 (6.9)
Athetoid	4 (3.1)
Motor distribution (n, %)	
One limb	2 (1.6)
Two limbs	67 (51.5)
Three limbs	13 (10.0)
Four limbs	48 (36.9)
Tube fed (n, %)	
Partial	11 (8.4)
Complete	5 (3.9)
VFSS (any)	19 (14.6)
Within study period (18–36 months)	9 (6.9)

Key: GMFCS, gross motor function classification system; SD, standard deviation; TD, typical development; VFSS, videofluoroscopic swallow study.

cough, choke, laboured breathing and colour change. The signs with the lowest agreement between raters (less than 85%) were gurgly voice, snuffly nose and multiple swallows.

4.3. Prevalence of clinical signs suggestive of pharyngeal phase impairments, and relationship to gross motor function

Overall, 67.7% of children with CP had clinical signs, rated by the clinician (live and/or from video) in a single mealtime. There was a stepwise increase in the proportion of children with clinical signs for each increase in level of gross motor function, as shown in Table 3. This was only significant for those in GMFCS IV and V (overall) when compared to the TD sample. This association with gross motor function was also noted for each individual clinical sign, except for cough, choke, throat clear, respiratory rate and snuffly nose. The most common signs on direct-assessment were coughing (44.7%),

Table 2
Reproducibility (inter-rater and intra-rater) of clinical signs suggestive of pharyngeal phase impairment in preschool children with cerebral palsy.

	Inter-rater ($n = 40$)		Intra-rater ($n = 40$)	
	Reliability (κ)	Agreement (%)	Reliability (κ)	Agreement (%)
Overall	0.7*	90.0	0.9*	95.0
Gag	0.6*	90.0	0.9*	97.5
Cough	0.9*	92.5	1.0*	100.0
Choke	0.4*	92.5	0.7*	97.5
Vomit	NC	NC	NC	NC
Throat clear	0.5*	95.0	−0.3	92.5
Multiple swallow	0.3*	80.0	0.5*	85.0
Wheeze	NC	NC	NC	NC
Stridor	NC	NC	NC	NC
Respiratory rate	NC	NC	0.0	97.5
Laboured breathing	0.7*	97.5	0.0	95.0
Wet breath	0.6*	85.0	0.7*	90.0
Gurgly voice	0.3*	77.5	0.5*	82.5
Snuffly nose	0.0	82.5	1.0*	100.0
Eye tearing	0.4*	87.5	−0.0	90.0
Colour change	0.4*	90.0	0.8*	97.5

* p value < 0.001. $\kappa < 0$ poor, 0.01–0.20 slight, 0.21–0.40 fair, 0.41–0.60 moderate, 0.61–0.80 substantial, and 0.81–1.0 almost perfect; NC due to too few rating categories.

Table 3
Clinical signs suggestive of pharyngeal phase impairment in preschool children with cerebral palsy, by GMFCS and food/fluid texture.

	TD: *n* (%) *N* = 40	I: *n* (%) *N* = 57	II: *n* (%) *N* = 15	III: *n* (%) *N* = 23	IV: *n* (%) *N* = 12	V: *n* (%) *N* = 23	OR (CI); *p*
Overall	15 (37.5)	32 (56.1)	9 (60.0)	15 (65.2)	9 (75.0)*	23 (100.0)*	1.7 (1.3, 2.1); <0.01
Cough[b]	15 (37.5)	26 (45.6)	4(26.7)	10 (43.5)	6 (50.0)	9 (56.3)	1.1 (0.9, 1.3); 0.32
Multiple swallow	0 (0.0)	1 (1.8)*	1 (6.7)*	7 (30.4)*	6 (50.0)*	16 (100.0)*	9.9 (3.9, 25.3); <0.01[a]
Gurgly voice	0 (0.0)	5 (8.8)*	1 (6.7)*	4 (17.4)*	5 (41.7)*	10 (62.5)*	2.5 (1.8, 3.6); <0.01[b]
Wet breath	3 (7.5)	8 (14.0)	2 (13.3)	3 (13.0)	2 (16.7)	8 (50.0)*	1.5 (1.1, 1.9); <0.01
Gag	0 (0.0)	3 (5.3)*	1 (6.7)*	1 (4.3)*	1 (8.3)*	8 (50.0)*	2.3 (1.5, 3.4); <0.01
Rattly chest	0 (0.0)	2 (4.1)*	1 (7.1)*	1 (5.0)*	1 (11.1)*	4 (36.4)*	2.1 (1.3, 3.4); <0.01
Respiratory effort	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	5 (31.3)*	38.7 (3.6, inf); <0.01[c]
Choke	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (8.3)	1 (6.3)*	3.5 (0.7, 16.2); 0.12
Throat clear	0 (0.0)	2 (3.5)*	0 (0.0)	2 (8.7)*	1 (8.3)	0 (0.0)	1.2 (0.7, 2.1); 0.41
Respiratory rate	0 (0.0)	1 (1.8)*	0 (0.0)	0 (0.0)	0 (0.0)	2 (12.5)*	2.1 (0.9, 4.5); 0.07
Snuffly nose	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (8.3)	2 (12.5)*	5.0 (1.0, 25.9); 0.06
Eye tearing	0 (0.0)	1 (1.8)*	0 (0.0)	0 (0.0)	0 (0.0)	2 (12.5)*	2.3 (1.0, 5.2); 0.05
Colour change	0 (0.0)	0 (0.0)	0 (0.0)	1 (4.3)*	1 (8.3)*	5 (31.3)*	5.1 (1.8, 14.3); <0.01

Vomit, stridor and wheeze not reported as not observed in the live ratings; seven children defaulted to impaired overall as nil by mouth; thin fluids $n = 119$ (12 defaulted to impaired, but no fluid rated), thick fluids $n = 11$ (7 defaulted to impaired), puree $n = 114$ (10 defaulted to impaired), semi-solid $n = 86$ (7 defaulted to impaired), chewable $n = 126$ (18 defaulted to impaired); inf, infinity; NA, not applicable, as no children on thickened fluids from GMFCS level; NC, not calculable; TD, typical development.

[a] Gender significantly related.
[b] Age significantly related.
[c] Calculated using episheet for GMFCS V compared to TD only.
* Indicates significantly greater proportion in GMFCS level compared to children with TD ($p < 0.05$).

multiple swallows (25.2%), gurgly voice (20.3%), wet breathing (18.7%) and gagging (11.4%) (Fig. 2). Many of the signs were more commonly observed on fluids, although multiple swallows and gag were more common on solid foods (Supplementary 1). Using the modified OPD cut-point (based on the validation with the TD sample), a more conservative prevalence estimate of 50.8% was found (TD = 12.5%, GMFCS I = 35.1%, II = 13.3%, III = 56.5%, IV = 66.7%, V = 100.0%).

Supplementary 1 related to this article can be found, in the online version, at http://dx.doi.org/10.1016/j.ridd.2014.12.021.

Parents reported clinical signs suggestive of pharyngeal phase impairment during mealtimes in 46.2% of their children. The proportion of children with signs increased with poorer gross motor function (GMFCS I: 36.8%, II: 40.0%, III: 43.5%, IV: 58.3%, V: 69.6%). The association was significant for the overall model (OR = 1.4, $p = 0.08$), however only significant for level V compared to level I (OR = 3.9, $p = 0.01$). The most common signs based on parent-report were coughing (30.5%), gagging

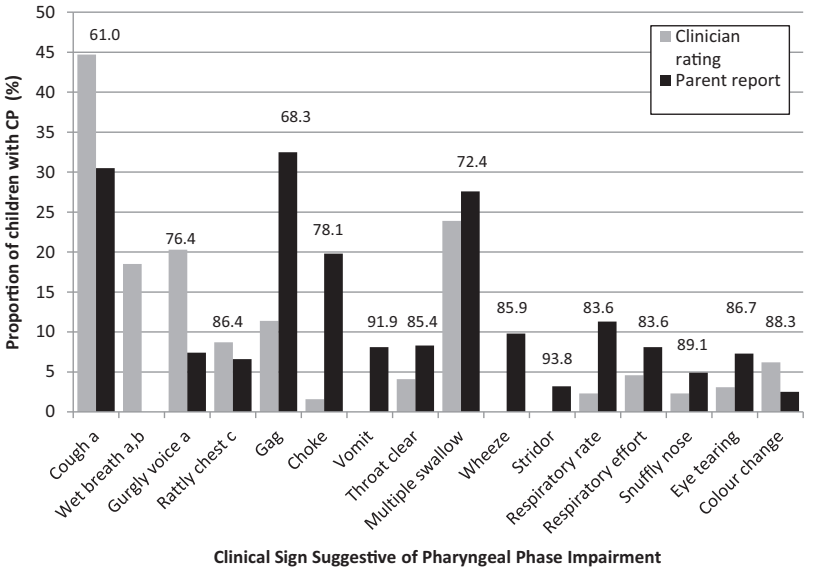

Fig. 2. Percentage agreement for clinical signs suggestive of pharyngeal phase impairment, based on direct ratings and parent report.
Percentage agreement between clinician rating and parent report indicated above bars; [a]Clinician rating completed live and confirmed from video; [b]Wet breathing was not included in the CP Child Feeding Questionnaire (parent-report); [c]Clinician rating completed live only (detected through touch).

(31.3%), choking (18.8%), and multiple swallows (25.2%) (Fig. 2). The parent-report agreed with direct-assessment in 60% of cases (kappa = 0.2, $p < 0.01$, Fig. 2). The mean of differences between the average number of signs per texture was 0.3 (SD = 2.2) suggesting no bias in parents over/under-reporting.

5. Discussion

This study is the first to our knowledge to report a representative population-based estimate of the prevalence of clinical signs suggestive of pharyngeal dysphagia in young children with CP. In particular, the discriminative validation of these signs in a sample of children with typical development, and testing of their reproducibility, gives greater confidence in the interpretation of the findings.

Clinical signs suggestive of pharyngeal dysphagia were observed in over a third of children aged 18–36 months with typical development. Only two signs were observed as part of typical development; coughing and wet breathing. In most cases, only a single cough on thin fluids was noted, and as such, this met our a priori criteria for excluding this sign in our definition of possible pharyngeal dysphagia. This study was focusing on the clinical observation of signs suggestive of pharyngeal phase impairment, thus we are unable to differentiate the causation of the observed sign (developmental, structural, physiological or neurological), and whether this varied between the TD and CP samples. In order to retain the naturalistic component of the mealtime, the volume and means of intake were not standardised between children. Thus, the increased coughing observed, particularly in the TD sample and in those with ambulatory CP, may also be associated with the introduction of more challenging fluid utensils (with less controlled flow rates and volumes, such as open cups and straws) and children's initiation of consecutive fluid swallows. DeMatteo and colleagues explored the diagnostic accuracy of clinical signs, proposing that predictive clusters of signs, such as a cough combined with voice changes and gag, was most predictive of fluid aspiration (DeMatteo et al., 2005). Thus a single cough, particularly on thin fluids, may not have sufficient discriminative validity to suggest pharyngeal dysphagia, particularly if only a single mealtime is observed.

Overall, the reproducibility of the clinical signs was strong, both with repeated ratings by one clinician, and between clinicians. As expected, signs which were more overt, such as coughing, choking and gagging, had the strongest reproducibility. Signs which were detected through more subtle perceptual changes, such as wet breathing, gurgly voice, eye tearing, snuffly nose, had lower reproducibility, particularly between clinicians. The presence of a cough, being among the most overtly observable signs, was the most reliably identified clinical sign by clinicians (almost perfect).

Clinical signs suggestive of pharyngeal phase impairment were common in 68% of preschool-aged children with CP. These findings were similar to those of Del Giudice, who studied clinical signs based on parent-report in children with CP (mean 5.2 years) (Del Giudice et al., 1999), although our estimate based on parent-report was lower. We found that there was a stepwise increase in proportion of children with clinical signs with each increase in GMFCS level, consistent with the broader literature on OPD (Benfer et al., 2013; Calis et al., 2008; Fung et al., 2002; Parkes et al., 2010; Reilly et al., 1996; Sullivan et al., 2000; Waterman et al., 1992). A surprising finding was the notable proportion of children from GMFCS I and II with clinical signs, even after applying the modified cut-points from the validation study (35.1 and 13.3%, respectively). Little has been reported on children with ambulatory CP in the literature with regards to clinical signs, so further investigation of this sub-group is warranted. Children with non-ambulatory CP almost consistently demonstrated clinical signs (in 91% of GMFCS IV and V). Previous studies have generally used indirect report of clinical signs or have not described their findings according to GMFCS, which reduces our ability to compare with our data. Only one study, by Calis and colleagues, used direct-assessment of clinical signs on the Dysphagia Disorders Survey, finding an equivalent proportion of children from GMFCS IV and V (91%) showed signs (Calis et al., 2008).

Coughing, multiple swallows, gurgly voice, wet breathing and gagging were the most commonly observed signs (>10% of children), with few children showing evidence of the other signs in a single mealtime. There were similar proportions of children demonstrating coughing during mealtimes in the current study compared to two other studies of preschool children with CP, although the gross motor severity of these samples was not well defined (Clancy & Hustad, 2011; Wilson & Hustad, 2009). In a third study of preschool children, by Reilly et al., the estimate was higher (70%), but included coughing and choking combined (Reilly et al., 1996). As Clancy and Hustad described, children with CP, even those assessed to have normal oromotor skills, may continue to demonstrate coughing during mealtimes until six years of age, suggesting a later maturation compared to children with TD (Clancy & Hustad, 2011). Fewer children from GMFCS V coughed on thin fluids compared to other GMFCS levels, which may reflect the findings in previous studies showing high rates of silent aspiration in children with more significant neurological lesions (Arvedson et al., 1994). This difference may also be attributable to children from GMFCS IV and V having more controlled fluid intake (due to modified utensils such as infant bottles and trainer cups, smaller volumes or single sips, or bolus pacing by the feeder).

This study showed that when parents were asked to observe specific clinical signs which were described in written form, their report agreed with direct-assessment in about 60% of the cases. This was not biased to consistently over- or under-report the number of signs compared to direct-assessment. The discrepancy in agreement may arise from differences in the number of mealtimes observed, the quantity and type of textures included (being a restricted range of textures in the standardised assessment), and the mealtime context. Interestingly, agreement was lower for the signs for which we would expect better accuracy by parents, such as coughing and gagging, being more overtly observable and

everyday familiar terms. Conversely, parents reported a surprisingly high prevalence of signs such as wheezing, stridor, respiratory rate and effort, vomiting and snuffly nose, which were almost non-existent in the direct-assessments. This may be related again to the duration and method of direct-assessment (video rating in a single mealtime), but may also reflect lack of clarity for parents surrounding some terms (Mellis, 2009). A cough was more commonly noted by clinicians than parents. Of those children observed to cough clinically but whose parents indicated their child does not cough, 81% had mild OPD. We hypothesise thus that the parent-report may be giving a more accurate reflection of children who cough regularly at mealtimes (which may be a better indicator of pharyngeal dysphagia), whereas the observation in a single mealtime may detect cases of isolated coughing. Parents were able to report on signs over a number of mealtimes, which meant their estimates may be more accurate in instances, as long as the term is clearly understood.

5.1. Limitations

This study has provided valuable data to fill a significant research gap, but had some key limitations. The study would have been strengthened by including an instrumental assessment for all children displaying any clinical signs (beyond a single cough on thin fluids). We reviewed data from videofluoroscopies performed as part of the children's standard clinical management, however only nine children had VFSS during the study period, which was insufficient for analysis. Without instrumental data, we are only able to comment on the presence of signs rather than infer impairment.

The location of neurological lesion influences the motor type which characterises an individual's CP. This may also influence the patterns of clinical signs observed during mealtimes. While this is an important interaction to be aware of in the field of OPD, the small numbers of the non-spastic motor types which are present in a representative population-based sample (i.e. dyskinetic, ataxic and hypotonic) were insufficient for statistical analysis of this relationship.

This study was conducted longitudinally over a 4 year period, and as such live clinical assessment by a speech pathologist was not feasible. Many of the clinical signs may be more accurately detected live, and better determined across a number of mealtimes, which may result in under-reporting in the direct-assessment.

6. Conclusions

This study has contributed to our understanding of which signs are most valid and reliable when applied to preschool children with CP. All 16 clinical signs used in this study had strong reproducibility by clinicians, suggesting they may be useful in the clinical setting, although further testing may be required to strengthen use in research. Exploring the test-retest reproducibility of these signs would assist in determining which are consistent between mealtimes and which are subject to greater variation. Cough was consistently identified by clinicians, but may not adequately reflect the child's performance across a number of mealtimes. The single cough on thin fluids was also common in children with TD, suggesting an isolated cough on thin fluids is not sufficiently discriminative in children aged 18–36 months. Considering coughing over a number of mealtimes, on multiple textures, or clustered with other signs may be a more appropriate marker in this age range. This study found clinical signs suggestive of pharyngeal phase impairments to be common in approximately 68% of children with CP, and this was present across all levels of gross motor function, including those with ambulatory CP. It is important for clinicians working in this field to be actively monitoring these signs, particularly as the available standardised OPD assessments focus on the oral phase and are not designed to assess the pharyngeal phase in adequate detail. The use of parent-report in both clinical screening and research studies remains a feasible method. Training parents to detect medical terms, such as stridor, wheeze, and rattly chest, and development of online training resources such as video-clips, may make this modality of screening more viable (Mellis, 2009). While studies analyzing the diagnostic accuracy of these signs related to aspiration on instrumental evaluation are available for the paediatric population, more studies of this kind are needed, particularly specific to CP.

Competing interests

The authors declare they have no competing interests.

Acknowledgements

We would like to thank Physiotherapists Rachel Jordan (BPT) and Chris Finn (BPT) for data collection and gross motor ratings; and Dietitians Joanne McMah (M Nutr & Diet), Stina Oftedal (B.Hlth.Sc (Hons) Nutr & Diet) and Camilla Davenport (B.Hlth.Sc (Hons) Nutr & Diet) for data collection of feeding videos. This project was supported by the National Health and Medical Research Council Postgraduate Medical and Dental Scholarship (1018264–KB), Career Development Fellowship (APP1037220–RB) and Project Grants (569605 and 465128). Funding was also received from the Speech Pathology Australia Post Graduate Student Research Grant to conduct reliability ratings.

Appendix A. Clinical signs with descriptions provided to parents on the Queensland Cerebral Palsy Child Feeding Questionnaire

10. Summary of my child's signs and symptoms during eating or drinking. These are a list of signs and symptoms of swallowing difficulty that your child may demonstrate during eating or drinking.

Place a tick (✓) in the box if you see your child doing this behaviour during eating or drinking for each consistency.

Signs or Symptoms	Does this happen at all?			If you ticked 'Yes', indicate (✓) the types of drinks or food textures on which your child demonstrates these signs or symptoms.				
My child.	I don't know	No	Yes	Thin drink	Thick drink	Smooth puree	Lumpy semi-solid	Finger foods
Gags when eating or drinking.								
Coughs when eating or drinking.								
Child chokes when eating or drinking.								
Vomits when eating or drinking.								
Clears his/her throat often during or after meals.								
Needs to swallow a number of times to clear each mouthful of food or drink.								
Wheezes during/after eating or drinking. (Wheezing is a whistling sound from the chest during breathing).								
Has 'stridor' when breathing in or out during eating or drinking. (Stridor is a harsh, high-pitched, vibratory noise in the throat particularly when breathing in.)								
Becomes breathless and breathes quickly during eating or drinking.								
Breathing becomes laboured or effortful during eating or drinking.								
Has a 'rattly chest' after eating or drinking.								
Gets a 'snuffly nose' after eating or drinking.								
Has a 'gurgly voice' after eating or drinking.								
Has runny eyes or 'eye tearing' after swallows of certain food or drinks.								
Seems to go 'blue' around the lips/face or turn 'dusky' or pale after drinking or eating.								
Regularly gets high temperatures.								
Generally refuses to eat or drink some food or fluid textures.								

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Supplementary Information 1: All clinical signs suggestive of pharyngeal phase impairment by texture and GMFCS

	TD: n (%) N=40	I: n (%) N=57	II: n (%) N=15	III: n (%) N=23	IV: n (%) N=12	V: n (%) N=23	OR (CI); p
Overall	16 (40.0)	32 (56.1)	9 (60.0)	15 (65.2)	9 (75.0)*	23 (100.0)*	1.7 (1.3,2.1); <0.01
Thin fluid	15 (37.5)	19 (34.6)	6 (42.9)	5 (29.4)	3 (27.3)	20 (90.9)*	1.4 (1.1,1.7); <0.01
Thick fluid	NA	NA	NA	NA	NA	11 (100.0)	NC [a]
Puree	3 (7.5)	4 (10.6)	1 (6.7)	4 (19.1)	5 (50.0)*	21 (100.0)*	2.8 (2.0,3.8); <0.01
Semi-solid	0 (0.0)	4 (11.4)*	0 (0.0)	4 (23.5)*	5 (62.5)*	15 (100.0)*	4.3 (2.5,7.5); <0.01
Chewable	3 (7.5)	9 (16.1)	2 (13.3)	11 (50.0)*	7 (58.3)*	21 (100.0)*	3.0 (2.2,4.1); <0.01[c]
Cough[b]	15 (37.5)	26 (45.6)	4(26.7)	10 (43.5)	6 (50.0)	9 (56.3)	1.1 (0.9,1.3); 0.32
Thin fluid	13 (32.5)	14 (25.0)	3 (21.4)	4 (23.5)	4 (36.4)	2 (22.2)	0.9 (0.7,1.2); 0.61
Thick fluid	NA	NA	NA	NA	NA	2 (50.0)	NC [a]
Puree	2 (5.1)	4 (8.5)	1 (7.1)	1 (4.8)	0 (0.0)	1 (7.7)	0.9 (0.6,1.5); 0.77
Semi-solid	0 (0.0)	2 (5.6)*	1 (9.1)*	2 (11.8)*	2 (28.6)*	1 (10.0)*	1.7 (1.1,2.6); 0.02
Chewable	3 (7.5)	7 (12.7)	2 (13.3)	3 (14.3)	1 (12.5)	5 (55.6)*	1.5 (1.1,2.1); 0.01[c]
Multiple swallow	0 (0.0)	1 (1.8)*	1 (6.7)*	7 (30.4)*	6 (50.0)*	16 (100.0)*	9.9 (3.9,25.3); <0.01[d]
Thin fluid	0 (0.0)	1 (1.8)*	0 (0.0)	0 (0.0)	2 (18.2)*	4 (44.4)*	3.5 (1.6,7.5); <0.01
Thick fluid	NA	NA	NA	NA	NA	NA	NC [a]
Puree	0 (0.0)	1 (2.1)*	1 (7.1)*	4 (19.1)*	4 (44.4)*	12 (92.3)*	6.7 (3.0,15.0); <0.01
Semi-solid	0 (0.0)	1 (2.8)*	0 (0.0)	3 (17.7)*	5 (71.4)*	9 (90.0)*	9.4 (3.0,29.5); <0.01
Chewable	0 (0.0)	1 (1.8)*	1 (6.7)*	6 (28.6)*	3 (37.5)*	9 (100.0)*	7.8 (3.2,19.1); <0.01[d]
Gurgly voice[b]	0 (0.0)	5 (8.8)*	1 (6.7)*	4 (17.4)*	5 (41.7)*	10 (62.5)*	2.5 (1.8,3.6); <0.01[c]
Thin fluid	0 (0.0)	2 (3.6)*	0 (0.0)	1 (5.9)*	1 (9.1)*	2 (22.2)*	1.9 (1.1,3.3); 0.02
Thick fluid	NA	NA	NA	NA	NA	2 (50.0)	NC [a]
Puree	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	4 (30.8)*	37.4 (2.2,inf); <0.01[e]
Semi-solid	0 (0.0)	1 (2.8)	0 (0.0)	0 (0.0)	0 (0.0)	2 (20.0)*	2.2 (1.0,4.9); 0.05
Chewable	0 (0.0)	0 (0.0)	0 (0.0)	1 (4.8)*	0 (0.0)	3 (33.3)*	11.5 (1.4,94.0); 0.02
Wet breath[b]	3 (7.5)	8 (14.0)	2 (13.3)	3 (13.0)	2 (16.7)	8 (50.0)*	1.5 (1.1,1.9); <0.01
Thin fluid	2 (5.0)	2 (3.6)	1 (7.1)	0 (0.0)	0 (0.0)	0 (0.0)	0.6 (0.3,1.5); 0.28
Thick fluid	NA	NA	NA	NA	NA	0 (0.0)	NC [a]
Puree	1 (2.6)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (7.7)	1.3 (0.6,2.9); 0.48
Semi-solid	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (10.0)	1.5 (0.6,4.4); 0.41
Chewable	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (12.5)	1 (11.1)	5.0 (0.4,57.7); 0.20
Gag	0 (0.0)	3 (5.3)*	1 (6.7)*	1 (4.3)*	1 (8.3)*	8 (50.0)*	2.3 (1.5,3.4); <0.01
Thin fluid	0 (0.0)	1 (1.8)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0.7 (0.1,4.5); 0.71
Thick fluid	NA	NA	NA	NA	NA	2 (50.0)	NC [a]
Puree	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	4 (30.8)*	0.2 (0.0,2.4); 0.19
Semi-solid	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	NC
Chewable	0 (0.0)	2 (3.6)*	1 (6.7)*	0 (0.0)	1 (12.5)*	4 (45.4)*	2.3 (1.4,4.0); <0.01
Rattly chest	0 (0.0)	2 (4.1)*	1 (7.1)*	1 (5.0)*	1 (11.1)*	4 (36.4)*	2.1 (1.3, 3.4); <0.01
Respiratory effort	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	5 (31.3)*	38.7 (3.6,inf); <0.01[e]
Thin fluid	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	3 (33.3)*	43.6 (3.4,inf); <0.01[e]
Thick fluid	NA	NA	NA	NA	NA	1 (25.0)	NC [a]
Puree	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	2 (15.4)	17.1 (0.6,inf);0.13[e]
Semi-solid	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	NC
Chewable	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (11.1)	14.3 (0.8,inf); 0.07[e]
Choke	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (8.3)	1 (6.3)*	3.5 (0.7,16.2); 0.12
Thin fluid	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	NC
Thick fluid	NA	NA	NA	NA	NA	1 (25.0)	NC [a]
Puree	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	NC
Semi-solid	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	NC
Chewable	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (12.5)	0 (0.0)	2.3 (0.6,9.6); 0.25
Throat clear	0 (0.0)	2 (3.5)*	0 (0.0)	2 (8.7)*	1 (8.3)	0 (0.0)	1.2 (0.7,2.1); 0.41
Thin fluid	0 (0.0)	2 (3.6)*	0 (0.0)	1 (5.9)*	1 (9.1)	0 (0.0)	1.3 (0.7,2.2); 0.44
Thick fluid	NA	NA	NA	NA	NA	0 (0.0)	NC [a]

Puree	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	NC
Semi-solid	0 (0.0)	0 (0.0)	0 (0.0)	1 (5.9)	0 (0.0)	0 (0.0)	1.7 (0.5,5.7); 0.42
Chewable	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	NC
Respiratory rate	0 (0.0)	1 (1.8)*	0 (0.0)	0 (0.0)	0 (0.0)	2 (12.5)*	2.1 (0.9,4.5); 0.07
Thin fluid	0 (0.0)	1 (1.8)*	0 (0.0)	0 (0.0)	0 (0.0)	2 (22.2)*	2.4 (1.0,5.6); 0.04
Thick fluid	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	NC [a]
Puree	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (7.7)	9.5 (0.6,inf); 0.13[e]
Semi-solid	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	NC
Chewable	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	NC
Snuffly nose	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (8.3)	2 (12.5)*	5.0 (1.0,25.9); 0.06
Thin fluid	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (11.1)	14.3 (0.8,inf);0.07 [e]
Thick fluid	NA	NA	NA	NA	NA	1 (25.0)	NC [a]
Puree	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	NC
Semi-solid	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	NC
Chewable	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (12.5)	0 (0.0)	2.3 (0.6,9.6); 0.25
Eye tearing	0 (0.0)	1 (1.8)*	0 (0.0)	0 (0.0)	0 (0.0)	2 (12.5)*	2.3 (1.0,5.2); 0.05
Thin fluid	0 (0.0)	1 (1.8)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0.7 (0.1,4.2); 0.70
Thick fluid	NA	NA	NA	NA	NA	2 (50.0)	NC [a]
Puree	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (7.7)	9.5 (0.6,inf); 0.13[e]
Semi-solid	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	NC
Chewable	0 (0.0)	1 (1.8)	0 (0.0)	0 (0.0)	0 (0.0)	1 (11.1)	2.1 (0.8,5.6); 0.13
Colour change	0 (0.0)	0 (0.0)	0 (0.0)	1 (4.3)*	1 (8.3)*	5 (31.3)*	5.1 (1.8,14.3); <0.01
Thin fluid	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (11.1)	14.3 (0.8,inf);0.07 [e]
Thick fluid	NA	NA	NA	NA	NA	2 (50.0)	NC [a]
Puree	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (7.7)	9.5 (0.6,inf); 0.13[e]
Semi-solid	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (14.3)	0 (0.0)	1.9 (0.5,7.1); 0.34
Chewable	0 (0.0)	0 (0.0)	0 (0.0)	1 (4.8)*	1 (12.5)*	3 (30.0)*	4.2 (1.5,11.7);<0.01

[a]All children on thickened fluids from GMFCS V, therefore association not calculable (NC); [b]Clinical sign rated live (not according to texture) and from video, therefore total with sign may not reflect sum of each texture with sign; [c]Age significantly related; [d]Gender significantly related; [e] Calculated using episheet for GMFCS V compared to TD only; Vomit, stridor and wheeze not reported as not observed in the live ratings; seven children defaulted to impaired overall as nil by mouth; thin fluids n=119 (12 defaulted to impaired, but no fluid rated), thick fluids n=11 (7 defaulted to impaired), puree n=114 (10 defaulted to impaired), semi-solid n=86 (7 defaulted to impaired), chewable n=126 (18 defaulted to impaired); inf infinity; NA Not applicable, as no children on thickened fluids from GMFCS level; NC Not calculable; TD Typical Development

Table 6. Results of Videofluorscopic Swallow Studies in Relation to Clinical Mealtime Assessment and Parent Reported Signs

Child	GMFCS	Time between VFSS & clinical assessment	Reason for VFSS referral	Foods at VFSS	Key findings of VFSS	Findings of clinical assessment	Parent report
1	I	3 months pre 10 months pre	OPD	All	Delayed trigger [all] Epiglottic undercoat [thk] P/A (incl silent) [thn] A (silent) [thk]	Nil	Nil
2	I	9 months pre	Discoordinated SSB	Puree Lumpy Thin/thick fluid	Delayed trigger [thn] Pooling in valleculae No P/A. Recommn: FO	Nil	Gag [pr, ss, chw] Cough [thn] Throat clear [pr] Multiple swallows [ss] Wheeze [pr] Respiratory effort [thn]
3	II	5 months pre	Cough/ choke on thin fluids	Puree Thin/thick fluid	Delayed and residue No p/a. Recommn: FO	Nil	Nil
4	III	2 months pre	Poor feeding, concern re aspiration	Puree Thin/thick fluid	A (trace, silent) [thn] Epiglottic undercoat [thk] Recommendation: Recommn: pr/ ss/ thk	Missing cgh post	Nil
5	V	4 months pre	Oral aversion, coughing on thin	Puree Thin/thick fluid	Pooling & delay A [thn] Delayed ineffective cough Recommn: pr/ thk	Gag [pr] Multiple swallows [pr] Wet breath [pr] Gurgly voice [pr]	Gag [thk, pr] Cough [thn, pr] Wheeze [pr]
6	V	2 months pre 1 month post	Queried aspiration	Puree Thick fluid	Delayed trigger P/A (incl silent) [pr]	Gag [thk, pr, ss] Cough [thk, chw] Multiple swallows [thk, ss, chw] Eye tearing [thk, chw] Colour change [thk]	Gag [thn, ss, chw] Cough [thn, ss, chw] Throat clear [thk, pr] Multiple swallows [pr] Wheeze [thk, pr] Fremitus [thk, pr]

7	V	8 months pre (NBM)			Pooling [thn/thk] Delayed trigger No P/A	NBM	Nil
8	V	7 months pre (NBM)	Poor feeding, reflux, chest infections. PEG fed with oral meals: assess safety of oral.	Puree	Delayed No P/A Recommn: purees only	NBM	NBM
9	V	2 months post		Puree Lumpy Thick fluid	Delay [all] A [thk]	Gag [chw] Multiple swallows [thk] Wet voice [thk] Wet respiration [ss, chw]	Multiple swallows [thk, pr, ss, chw]

Abbreviations: A aspiration; chw chewable; FO full oral; GMFCS Gross Motor Function Classification System; NBM nil by mouth; P penetration; PEG percutaneous endoscopic gastrostomy; pr puree; recommn recommendation; ss semi-solid; thk thick fluid; thn thin fluid; VFSS videofluoroscopic swallow study

Summary of Chapter 8

This chapter provided an understanding of the prevalence and patterns of clinical signs associated with pharyngeal phase impairment common in preschool children with CP, and how these vary by level of gross motor function.

i. A single cough on thin fluids was commonly observed in children with TD (coughing was observed in 38% of sample). This was therefore considered to be part of typical development in children younger than 36 months, and excluded from the modified definition of possible pharyngeal dysphagia.
ii. All 16 signs used in the study had strong reproducibility by clinicians (>90% for overall pharyngeal dysphagia). Cough was most reliably identified by clinicians.
iii. The proportion of children with clinical signs suggestive of pharyngeal phase impairments was 68% (51% using modified cut-points). This was lower than the proportion of children with oral phase impairments.
iv. Impairments were present in children from all GMFCS levels, including those with ambulatory CP. GMFCS was strongly associated with impairments of the pharyngeal phase, with the proportion of children with clinical signs suggestive of pharyngeal phase impairment significantly greater for children in GMFCS IV-V compared to children with TD.
v. The clinical signs suggestive of pharyngeal phase impairment most frequently noted were coughing (45%), multiple swallows (24%), gurgly voice (20%), wet breathing (19%), and gagging (11%).
vi. Agreement between parent report and direct assessment was moderate (60%), and parents did not consistently overreport or underreport (based on the total number of clinical signs). Parent report can be performed over a number of mealtimes which may reduce detection of isolated signs which are occurring in a single mealtime only. This may be a feasible method of screening and monitoring change.
vii. It is important for clinicians working in the field of CP to be actively monitoring clinical signs suggestive of pharyngeal phase impairment, particularly as the available standardised OPD measures do not adequately assess this phase of feeding.

The preceding chapters have focused on understanding OPD in preschool children with CP in a series of cross-sectional studies. As the preschool years are a time of rapid development, it is important to understand how OPD progresses longitudinally.

Chapter 9: Longitudinal Changes in Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy

Introduction to Chapter 9

The manuscript “Longitudinal Changes in Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy” is presented in this chapter, and consists of the findings from substudy 3. It aimed to understand changes in OPD prevalence and severity between 2 critical ages, 18 to 24 months and 36 months ca, according to gross motor function on the GMFCS. This is a period of significant transition for children’s oropharyngeal sensorimotor skills, in which the range of textures, utensils and mealtime routines place additional burden on their system. In order to adequately interpret the findings of this study, it was first necessary to test the test-retest reproducibility of the OPD measures. This substudy also explored risk factors for OPD at each assessment point, and the association between OPD at 18 to 24 months and health outcomes at 36 months (nutritional status, nutritional interventions, respiratory health and parent stress). This will assist in earlier intervention by understanding children who are likely to have persisting OPD, and those who show maturation of their feeding skills. It will also contribute to clinicians’ understanding of those aspects of OPD that are related to important health outcomes.

Paper 8: Longitudinal Changes in Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy

This article was submitted to Research in Developmental Disabilities, and is currently under review.

Longitudinal Study of Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy

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What's Known on this Subject

OPD in children with CP has commonly persisted since infancy. There are minimal changes in OPD during the early school years for children with CP (between 4 and 7 years). The progression of OPD between infancy and school age has not been systematically investigated.

What this Study Adds

This was a longitudinal analysis of childhood OPD in CP, in contrast to cross-sectional studies. The proportion of children with CP and OPD remained stable between 18 to 24 and 36 months; although OPD severity reduced in 30%. Gross motor function was the strongest OPD risk factor. OPD was associated with poor nutritional status at 36 months.

Acknowledgements

We would like to thank Physiotherapists Rachel Jordan (BPT) and Chris Finn (BPT) for data collection and gross motor ratings; and Dietitians Stina Oftedal (B.Hlth.Sc (Hons) Nutr & Diet) and Camilla Davenport(B.Hlth.Sc (Hons) Nutr & Diet) for data collection of feeding videos. Ethics approvals were gained through the University of Queensland Medical Research Ethics Committee (2008002260), Children's Health Services District Ethics Committee (HREC/08/QRCH/112), and other regional/ organisational ethics committees.

Abstract

Objective: To determine changes in prevalence and severity of oropharyngeal dysphagia (OPD) in children with cerebral palsy (CP) and relationship to health outcomes.

Methods: 53 children with confirmed CP diagnosis participated in this longitudinal study, assessed first at 18 to 24 months (Ax1 mean age 22.9 months ca (SD=2.9), 33 males, Gross Motor Function Classification System (GMFCS) I=22, II=7, III=11, IV=5, V=8) and at 36 months (Ax2). OPD was classified using the Dysphagia Disorders Survey (DDS) and signs suggestive of pharyngeal dysphagia. Nutritional status was measured using z scores for weight, height, and body mass index (BMI). Gross motor skills were classified on GMFCS and motor type/ distribution.

Results: Prevalence of OPD reduced from 62% to 59% between ages. 30% of children had an improvement to severity of OPD (>smallest detectable change), and 4% had poorer OPD. Gross motor function was strongly associated with OPD at both assessments, on the DDS (Ax1 OR=20.3, *P* = .011; Ax2 OR=28.9, *P* = .002), pharyngeal signs (Ax 1 OR=10.6, *P* = .007; Ax2 OR=15.8, *P* = .003), and OPD severity (Ax1 β=6.1, *P* < .001; Ax2 β=5.5 *P* < .001). OPD at 18 to 24 months was related to health outcomes at 36 months: low z scores for weight (adj β=1.2, *P* = .03) and BMI (adj β=1.1, *P* = .048), increased parent stress (adj OR=1.1, *P* = .049).

Conclusions: Classification and severity of OPD remained relatively stable between 18 to 24 months and 36 months. Gross motor function was the best predictor of OPD. These findings contribute to developing more effective screening processes considering critical developmental transitions anticipated to present challenges for children from different GMFCS levels.

Introduction

Oropharyngeal dysphagia (OPD) is common in approximately 85% of preschool children with cerebral palsy (CP),[1] although this estimate may be lower when accounting for feeding limitations associated with typical development.[2] CP is a lifelong disability of central origin influencing motor control, including that needed for effective and efficient eating, drinking and saliva control.[3] A number of important health outcomes have been associated with OPD, such as restricted growth and nutrition, compromised respiratory health, and increased parental stress during mealtimes.[4-6]

Children's feeding skills typically undergo a series of important changes through the preschool years, from suckle-feeding in infancy, to the rapid oropharyngeal skill changes

and encephalization during transitional feeding (4 to 36 months), and finally a period of skill consolidation (3 to 6 years).[7-9] The range of food textures and fluid utensils children can safely, efficiently, and independently manage are gradually expanded, owing to a range of influences, particularly the development of children's oropharyngeal sensorimotor systems. By 18 to 24 months, children can typically ingest firm and dual-textured foods[9,10] and from 24 to 36 months they can regularly drink from an open cup.[10] These periods of feeding development may present varied challenges for children with CP, as more complex textures, greater volumes of intake, more challenging utensils and increased mealtime independence/ routines place additional requirements on their oral sensorimotor, swallow-respiratory and cognitive systems.

Previous research supports the supposition that much of the OPD in children with CP has persisted since infancy, including reports of early difficulties with sucking, swallowing, or transition to solid foods.[11-14] Despite this, OPD may emerge during childhood in children with normal feeding in infancy,[11] and those presenting with difficulties in infancy may proceed to have typical feeding in childhood.[14-16] There has been limited exploration of longitudinal changes to feeding during the preschool years in children with CP. The feeding skill progression of 23 children with CP was explored in a study by Clancy and colleagues, collecting information through parent report from 4 to 7 years.[17] This study found significant differences in the proportion of impaired feeding skills between OPD severity groups (except for coughing/ choking), but only coughing reduced longitudinally.[17] Clancy's study emphasised the need for longitudinal research in children younger than 4 years in order to facilitate earlier intervention. The aim of the present longitudinal study, therefore, was to explore change in OPD prevalence and patterns in children with CP between two critical time points, 18 to 24 months and 36 months. Further, we aimed to understand whether feeding at 18 to 24 months could predict health outcomes (nutritional, respiratory and parent stress) at 36 months. Before evaluating change in OPD classification and severity, the test-retest reproducibility of measures had to be established. It was hypothesised that children with ambulatory CP may have delayed feeding at 18 to 24 months, but by 36 months fewer children would be classified as having OPD.

Patients and Methods

This longitudinal cohort study of preschool-aged children with CP was conducted in Queensland, Australia between April 2009 and April 2013. It is part of 2 larger studies exploring relationships between growth, nutrition and physical activity[18] and brain structure

and motor function in children with CP.[19] All caregivers consented for their child to participate with relevant institutional ethics gained.[18-20]

Patients

Children with a confirmed diagnosis of CP, aged 18 to 24 months corrected age (ca) at initial assessment, and born in Queensland between 2006-2009, were invited to participate. Only children returning for assessment at 36 months ca were included in this paper. Children with neurodegenerative conditions were excluded.

Forty children participated in the reproducibility substudy, aged between 18 to 36 months ca and having a confirmed diagnosis of CP (n=4 per GMFCS level per age band, stratified to 18 to 24 months and 30 to 36 months). This sample was recruited primarily through the main study sample, and additional children recruited through the CP Health Service, Royal Children's Hospital, Brisbane.

Measures

Measures of Oropharyngeal Dysphagia

Three standardised clinical measures of OPD were selected following systematic review of measure psychometrics (Dysphagia Disorders Survey -- Paediatric (DDS), Schedule for Oral Motor Assessment, and Pre Speech Assessment Scale.[20,21] A subsequent reproducibility and validity study resulted in the selection of the DDS with modified cut-points as the best available measure of OPD for research in preschool children with CP.[2] The DDS part 2 consists of a series of binary judgments of feeding competency on 8 ingestion functions for puree, chewable food and fluid, giving a maximum impairment raw score out of 22.[22]

Observation of 16 clinical signs suggestive of pharyngeal phase impairment was included in the determination of OPD classification, as the DDS provides insufficient detail on this phase of swallowing.[1] OPD classification was based on presence of 1 or more signs, with the exception of a single cough on thin fluids.[23]

Two secondary measures of OPD were included as early predictors of health outcomes. Feeding efficiency was calculated from average intake (grams) and time (minutes), recorded on a 3-day weighed diet record completed by parents at home.[18] Challenging behaviours demonstrated regularly during feeding (at least once daily) were reported by parents using the CP Child Feeding Questionnaire (CPFQ, Supplementary 1, Question 6). The total number of challenging behaviours, out of 16, was used to indicate possible sensory or behavioural feeding difficulties.

Risk Factors for Oropharyngeal Dysphagia

Children were classified on the Gross Motor Function Classification System (GMFCS) according to their age using the <2 years and 2-4 year age bands.[24] Motor type (spasticity, dyskinesia, hypotonia/ ataxia) and distribution (number of limbs) were also classified.[25,26]

Socioeconomic status (SES) was measured using the Socio-Economic Indexes for Areas (SEIFA), Index of Relative Socio-Economic Disadvantage[27] which assigns families to a decile rank (from 1= most disadvantaged to 10= least disadvantaged) based on family's postcode of residence. Preterm status was indicated for births with gestational age less than 37 weeks (time between first day of the last menstrual period and child's date of birth).[28] Presence of epilepsy was collected from parents during the initial physician interview.[19]

Measures of Health Outcomes

Nutritional status was indicated by gender- and age-referenced z scores for height, weight and body mass index (BMI).[29] Height or length (depending on children's ability to stand) was measured to the last completed millimetre by a length board (Shorr Productions, Maryland USA). Where direct measures of height/ length were not possible (due to contractures), height was estimated using published equations from knee length or upper-arm length[30] measured with an anthropometer (Holtain Ltd, UK). Weight was measured to the nearest 100 grams using chair scales (Seca, Germany), and BMI calculated as weight/ height (metres)^2.

Children's feeding method was reported by parents on a 5-point ordinal scale on the CPFQ (from total oral intake to total tube-feeding; SI1, Question 12). Parent stress associated with feeding their child was self-reported on a 5-point ordinal scale on the CPFQ (SI1, Question 7a). Respiratory illness was indicated by a hospitalisation for chest infection, diagnosis of pneumonia and/ or respiratory infection in the 6 months prior to assessment.[18-20]

Procedures

Children attended the hospital for anthropometry, mealtime and gross motor function assessments. During the mealtime assessment (video-taped for rating by a paediatric speech pathologist), 3 standardised presentations of 4 textures (puree, lumpy, chewable and fluid) were given by the carer, using their regular utensils.[31] Growth

anthropometry was measured by trained researchers, and gross motor function classifications conducted by 2 physiotherapists.

Reproducibility Substudy

For the test-retest reproducibility substudy, children were seen twice within a month for mealtime assessment. On both occasions the same procedures were followed and the same battery of tests conducted. The time, location and foods were kept as consistent as possible. Reproducibility was analysed using percentage agreement, kappas (binary) and Intra-Class Correlation Coefficients (ordinal scales >5 groups). The smallest detectable change (SDC) was calculated for the DDS raw score to determine score change that constituted true change in OPD (classification or severity). Clinical signs with agreement <80% were excluded from the definition of change for pharyngeal phase OPD.

Statistical Analysis

Participant characteristics, including OPD prevalence, were presented descriptively for both assessments, and change reported as a percentage and using McNemar's Test (binary), Wilcoxon Matched Pairs Test (ordinal) and paired *t* test (continuous). Potential OPD risk factors (age, gender, GMFCS (collapsed I-II, III, IV-V), BMI z score, preterm status, epilepsy, SES) were explored through mixed effects logistic regression for the presence of OPD outcomes (on the DDS and pharyngeal signs) and using mixed effects linear regression for OPD severity (DDS raw score). All models included *participant* as a random effect to account for within-participant dependence across the 2 assessment points, and *appointment* and *GMFCS* as interaction terms. First, univariate models were conducted, then multivariate models, using the above-listed risk factors as fixed effects. Association between OPD variables at 18 to 24 months and health outcomes at 36 months (nutritional status, introduction of supplementary feeding/ gastrostomy, parent stress, and hospitalisation for chest infection) were explored using logistic regression (binary outcomes) and linear regression (continuous outcomes). These models were adjusted for collapsed GMFCS at 36 months and gender. All analyses were performed using Stata 10.0 (Statacorp 2007), with significance set at $P < .05$.

Results

Sample Characteristics

There were 53 children who participated, aged 22.9 months (SD=2.9) at initial assessment (see Supplementary Information 2 for recruitment pathways and missing data). Sample

characteristics at each assessment and change between assessments are reported in Table 1. The sample's motor type distribution was not significantly different from the Australian CP Register at both assessments (Ax1: *P* = .81; Ax2: *P* = .37, χ^2 test), although GMFCS classification differed at the second assessment (Ax1: *P* = .09; Ax2: *P* = .001, χ^2 test).

Test-Retest Reproducibility

Reproducibility of the DDS overall was strong, and for clinical signs was moderate, as shown in Supplementary Information 2 (including data from SOMA and PSAS). Using the modified cut-points,[2] reproducibility for the DDS improved, with 90% agreement (κ=0.8, *P* < .001). The variability within the child's performance between mealtimes was greater than that attributable to intrarater variability[2] (Figure 2 for measurement error and SDC). Coughing was the most variable sign between mealtimes, with 60% agreement (κ=0.2, *P* = .10).

Prevalence of OPD

The prevalence of OPD reduced from 62% (n=33) at 18 to 24 months to 59% (n=31) at 36 months, as shown in Figure 1 (see SI2 for information on change based on the SOMA, PSAS and unmodified scoring). Four children changed from having OPD at 18 to 24 months to having no OPD at 36 months (all GMFCS I), and 2 children gained a classification of OPD at the second assessment (1 each from GMFCS I and III). Decline in OPD status was related to the presence of clinical signs suggestive of pharyngeal phase impairments at assessment 2.

The change in DDS scores overall, and on specific items (according to gross motor function) is shown in Figure 2 and 3, respectively. Fourteen children (30%) had an improvement in DDS score greater than that attributable to the test-retest SDC, and 2 children (4%) had a greater decline in scores. Only 1 child who was reclassified to poorer gross motor function had a decline in OPD, and 2 who declined in gross motor function improved in OPD classification.

Risk factors for oropharyngeal dysphagia and association with health outcomes

Gross motor function was the only risk factor for OPD that persisted between assessment 1 and 2 (Table 2). Age and epilepsy were also related to certain OPD outcomes and at certain assessment points. The relationship between OPD variables at 18 to 24 months and associated health outcomes at 36 months are reported in Table 3.

Table 1. Characteristics of Preschool-aged Children with Cerebral Palsy in the Longitudinal Oropharyngeal Dysphagia Study

	18 to 24 months n(%)	**36 months n(%)**	**Change n (%)[a]**	**Statistic (*P* value)[b,c,d]**
Gender, males:	33 (62%)	n/a	n/a	
GMFCS level:			10 (19%)	(1.00)[b]
I	22 (42%)	26 (49%)	0 (0%)	
II	7 (13%)	1 (2%)	6 (86%)	
III	11 (21%)	11 (21%)	2 (18%)	
IV	5 (9%)	7 (13%)	1 (20%)	
V	8 (15%)	8 (15%)	1 (13%)	
Primary motor type:			6 (11%)	(0.55)[b]
Spasticity	47 (88%)	47 (88)	5 (11%)	
Dyskinesia	2 (4%)	4 (8%)	0 (0%)	
Ataxia	3 (6%)	0 (0%)	3 (100%)	
Hypotonia	1 (2%)	2 (4%)	0 (0%)	
Motor distribution			5 (9%)	(0.18)[b]
Unilateral	18 (34%)	16 (30%)	2 (11%)	
Diplegia	10 (19%)	10 (19%)	2 (20%)	
Triplegia/ Quadraplegia	25 (47%)	27 (51%)	1 (4%)	
Preterm birth (<37 weeks)	28 (53%)	n/a	n/a	n/a
Epilepsy	9 (19.0%)	n/a	n/a	n/a
Socio-Economic Status (SEIFA)		n/a	n/a	n/a
Least disadvantaged (8-10)	14 (26%)			
Moderate disadvantage (5-7)	27 (51%)			
Most disadvantaged (1-4)	12 (23%)			
Tube/ supplementary feeding				(0.31)[b]
Full oral	34 (64%)	40 (75%)	3 (9%)	
Supplementary	15 (28%)	8 (15%)	10 (67%)	
Partial tube (mostly oral)	0 (0%)	0 (0%)	0 (0%)	
Partial tube (mostly tube)	3 (6%)	3 (6%)	1 (33%)	
Non-oral	1 (2%)	2 (4%)	0 (0%)	
Height for age Z score (mean, SD)	-0.9 (1.9)	-0.7 (1.2)	0.2	1.1 (0.28)[c]
Weight for age Z score (mean, SD)	-0.4 (1.6)	-0.6 (1.5)	-0.2	-1.9 (0.07)[c]
BMI Z score (mean, SD)	0.0 (1.9)	-0.2 (1.5)	-0.2	-0.7 (0.49)[c]
Respiratory illness				
Hospitalization for chest infection	4 (8%)	5 (9%)	7 (13%)	0.14 (0.71)[c]
Pneumonia	3 (6%)	2 (4%)	5 (10%)	0.2 (0.66)[d]
Respiratory infection	30 (57%)	23 (44%)	21 (40%)	2.3 (0.13)[d]

Abbreviations: BMI, Body Mass Index; GMFCS, Gross Motor Function Classification System; n/a, not applicable or available; SD, Standard Deviation; SEIFA, Socio-Economic Indexes for Areas

[a] Percentage change calculated based on number of children reclassified between assessment 1 and 2, divided by number in original group (assessment 1)
[b] Wilcoxon Matched Pairs Test.
[c] Paired t test.
[d] McNemars Test.

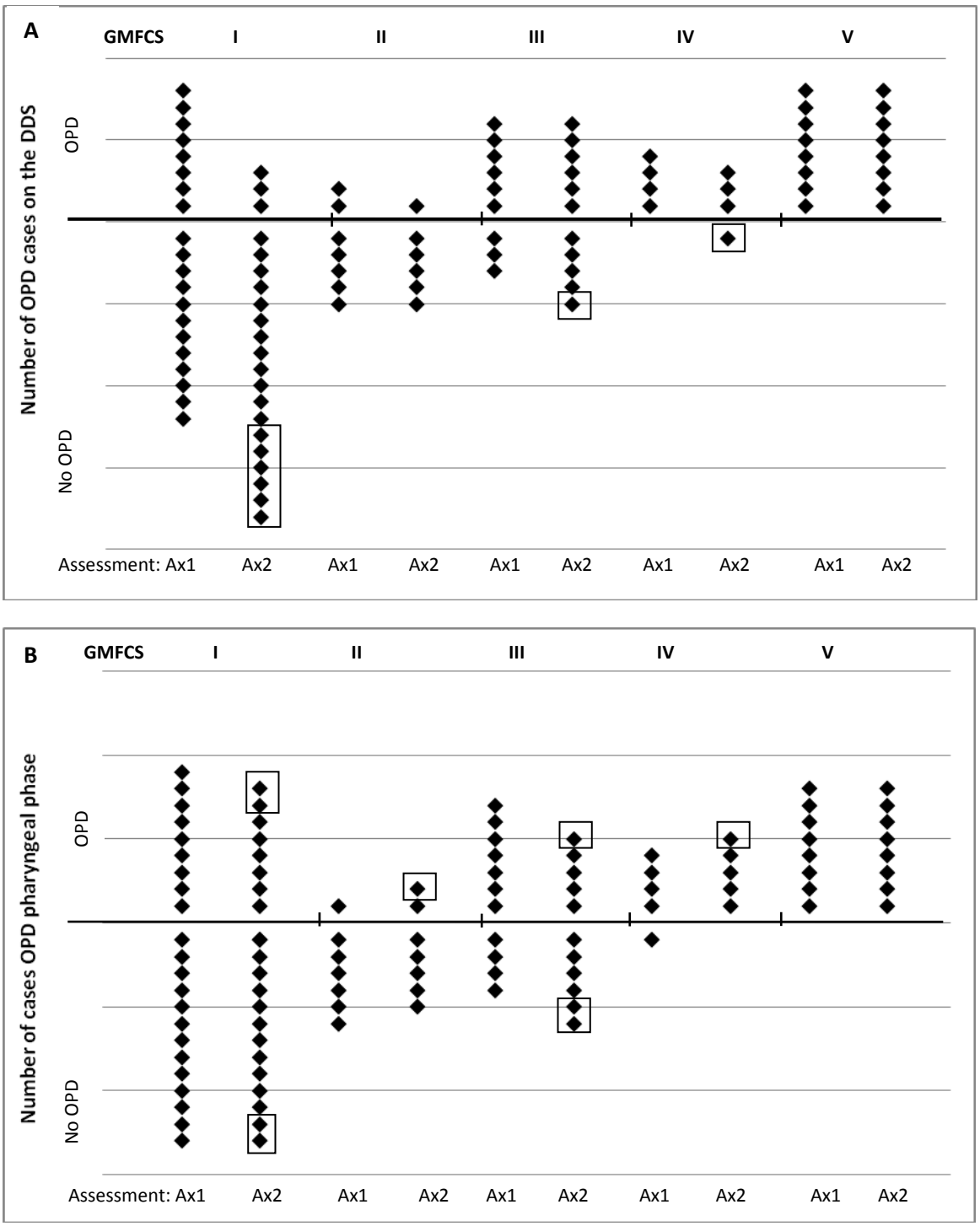

Figure 1. Change in Oropharyngeal Dysphagia Classification Between Assessment 1 (18 to 24 Months) and Assessment 2 (36 Months), According to Gross Motor Function (GMFCS) at Assessment 1

Abbreviations: Ax, Assessment; DDS, Dysphagia Disorders Survey; GMFCS, Gross Motor Function Classification System; OPD, Oropharyngeal Dysphagia

Fig. 1A OPD classification on DDS, Fig. 1B OPD classification on pharyngeal signs; Box indicates children reclassified; OPD classification based on modified cut-points[2]; Different numbers of children had a DDS score calculable between their 18 to 24 month assessment and 36 month assessment (n=1 GMFCS II, n=2 GMFCS III)

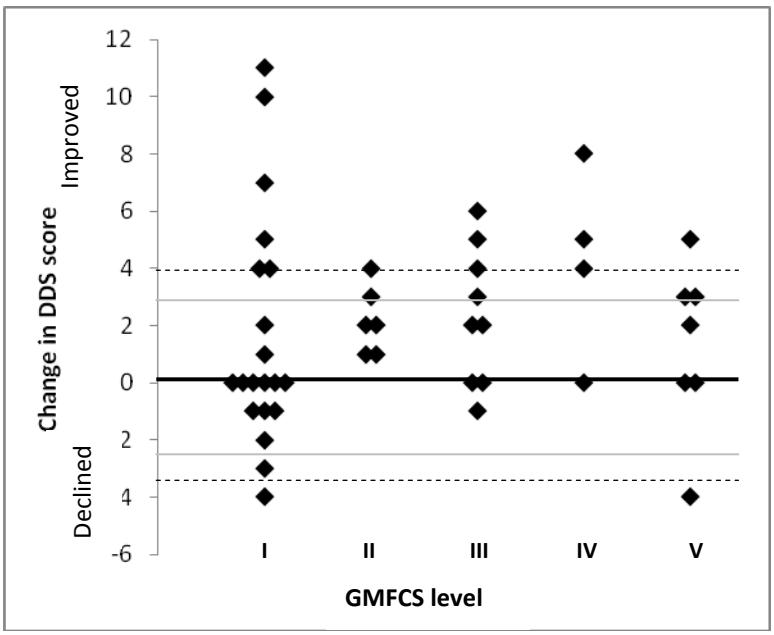

Figure 2. Change in Oropharyngeal Dysphagia Severity (Dysphagia Disorders Survey Raw Score) Between Assessment 1 (18 to 24 Months) and Assessment 2 (36 Months), According to Gross Motor Function (GMFCS) at Assessment 1

Abbreviations: DDS, Dysphagia Disorders Survey; GMFCS, Gross Motor Function Classification System

Dashed line indicates Smallest Detectable Change for test-retest (measurement error=1.4, smallest detectable change=3.8); solid line represents smallest detectable change for intra-rater,[2] (measurement error=1.0, smallest detectable change=2.8). Mean change for GMFCS I=1.6 (SD=4.1), II=2.2 (SD=1.2), III=2.3 (SD=2.4), IV=4.3 (SD=3.3), V=1.3 (SD=2.9), but these differences were not significant on linear regression (p=0.67).

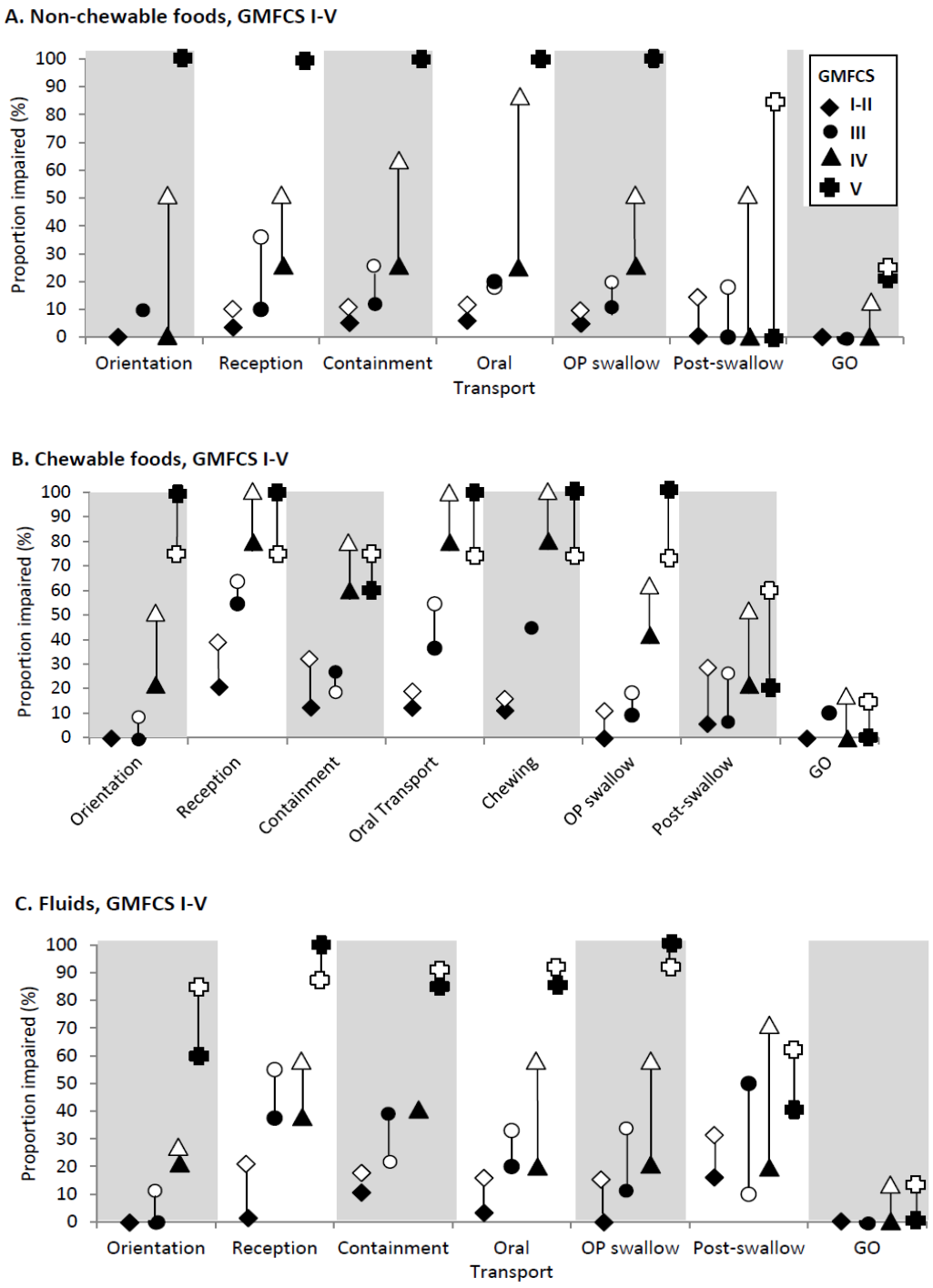

Figure 3. Proportion of Change in Ingestion Functions on the Dysphagia Disorders Survey Between Assessment 1 (18 to 24 Months) and Assessment 2 (36 Months), According to Gross Motor Function (GMFCS at 36 Months)

Abbreviations: OP, Oropharyngeal; GMFCS, Gross Motor Function Classification System; GO, Gastro-Oesophageal

Empty markers represent 18-24 month assessment, solid markers represent 36 month assessment, markers left to right for each ingestion function represent GMFCS I-V (GMFCS I-II combined as only n=1 in GMFCS II: I-II n=27, III n=11, IV n=7, V n=8); Difference in Dysphagia Disorders Survey texture scores between 18 to 24 months and 36 months based on paired *t* test: non-chewable score t=3.5, $P < .01$, chewable score t=4.2, $P < .01$, fluid score t=2.5, $P = .02$

Table 2. Comparison of Oropharyngeal Dysphagia Risk Factors at 18 to 24 Months ca (Assessment 1) and 36 Months ca (Assessment 2)

	Assessment at 18 to 24 months	**Assessment at 36 months**
OPD on DDS (modified)	**OR (95% CI); *P* value**	**OR (95% CI); *P* value**
GMFCS (collapsed)	20.3 (2.0, 208.4); 0.011	28.9 (3.4, 248.8); 0.002
I-II	ref	ref
III	8.1 (0.6, 117.7); 0.13	23.5 (1.3, 418.5); 0.032
IV-V	NC	NC
Motor type (collapsed)	NC	NC
Spasticity	ref	ref
Dyskinesia	2.3 (0.1, 69.6); 0.64	NC
Hypotonia/ ataxia	NC	NC
Body Mass Index z score	1.0 (0.5, 1.7); 0.89	0.7 (0.3, 1.5); 0.35
Preterm	0.4 (0.0, 6.4); 0.48	0.4 (0.2, 5.9); 0.49
Age	1.0 (0.7, 1.7); 0.86	0.2 (0.0, 1.3); 0.09
Gender (ref male)	0.8 (0.1, 14.9); 0.90	0.5 (0.0, 8.8); 0.66
Socio-economic status	1.0 (0.6, 1.8); 0.97	1.3 (0.7, 2.3); 0.46
Epilepsy	NC	NC
OPD severity (DDS raw score)	**β (95% CI); p value**	**β (95% CI); p value**
GMFCS (collapsed)	6.1 (4.6, 7.6); <0.001	5.5 (4.0, 7.0); <0.001
I-II	ref	ref
III	4.1 (1.0, 7.2); 0.009	2.5 (-0.6, 5.5); 0.11
IV-V	12.9 (10.0, 15.9); <0.001	11.7 (8.8, 14.5); <0.001
Motor type (collapsed)	0.3 (-1.6, 2.2); 0.77	1.7 (-0.4, 3.8); 0.10
Spasticity	ref	ref
Dyskinesia	1.2 (-4.7, 7.1); 0.69	2.7 (-1.6, 7.0); 0.22
Hypotonia/ ataxia	0.4 (-3.7, 4.6); 0.83	3.0 (-2.0, 8.0); 0.23
Body Mass Index z score	0.0 (-0.5, 0.5); 0.98	-0.4 (-1.0, 0.3); 0.24
Preterm	-0.4 (-4.4, 3.6); 0.86	-1.1 (-5.1, 2.9); 0.58
Age	-0.1 (-0.4, 0.3); 0.70	-0.78 (-1.9, 0.4); 0.18
Gender (ref male)	-0.8 (-5.0, 3.3); 0.70	-4.1 (-4.5, 3.7); 0.85
Socio-economic status	0.1 (-0.7, 0.9); 0.82	0.3 (-0.5, 1.1); 0.48
Epilepsy	8.1 (3.3, 12.8)); 0.001	7.5 (2.7, 12.2); 0.002
OPD of pharyngeal phase	**OR (95% CI); p value**	**OR (95% CI); p value**
GMFCS (collapsed)	10.6 (1.9, 59.2); 0.007	15.8 (2.5, 99.8); 0.003
I-II	ref	ref
III	8.6 (0.7, 105.3); 0.09	3.4 (0.3, 34.1); 0.31
IV-V	NC	NC
Motor type (collapsed)	1.9 (0.3, 11.1); 0.46	2.5 (0.3, 24.1); 0.42
Spasticity	ref	ref
Dyskinesia	NC	NC
Hypotonia/ ataxia	2.3 (0.1, 69.6); 0.64	1.3 (0.0, 176.6); 0.93
Body Mass Index z score	1.5 (0.8, 2.6); 0.18	1.0 (0.5, 2.0); 0.90
Preterm	0.8 (0.1, 7.3); 0.87	1.1 (0.1, 10.0); 0.91
Age	0.7 (0.4, 1.0); 0.06	0.4 (0.1, 1.5); 0.19
Gender (ref male)	2.0 (0.2, 19.1); 0.55	1.3 (0.1, 12.4); 0.81
Socio-economic status	1.2 (0.8, 1.9); 0.47	1.3 (0.8, 2.0); 0.31
Epilepsy	52.7 (1.1, 2433.9); 0.04	43.0 (1.0, 1901.6); 0.052

Abbreviations: BMI, Body Mass Index; DDS, Dysphagia Disorders Survey; GMFCS, Gross Motor Function Classification System; OPD, Oropharyngeal Dysphagia; NC, Not calculable as exposures predict outcome perfectly; OR, Odds Ratio; SEIFA, Socio-Economic Indexes for Areas

Table 3. Prediction of Health Outcomes in Children with Cerebral Palsy at 36 Months ca Based on Oropharyngeal Dysphagia at 18 to 24 Months ca

	Nutritional status					
	Height for Age z score		**Weight for Age z score**		**BMI z score**	
	Crude B (*P* value)	**Adjusted B (*P* value)[b]**	**Crude B (*P* value)**	**Adjusted B (*P* value)[b]**	**Crude B (*P* value)**	**Adjusted B (*P* value)[b]**
OPD on DDS	-0.3 (0.56)	0.01 (0.98)	0.4 (0.50)	0.8 (0.19)	0.9 (0.11)	1.1 (0.054)
Modified	-0.04 (0.92)	0.5 (0.26)	0.3 (0.50)	1.2 (0.03)[e]	0.5 (0.30)	1.1 (0.048)[e]
OPD severity[a]	-0.01 (0.64)	0.04 (0.29)	-0.03 (0.39)	0.03 (0.58)	-0.03 (0.34)	-0.01 (0.78)
Feeding efficiency	0.05 (0.36)	0.1 (0.36)	0.1 (0.11)	0.1 (0.09)	0.1 (0.10)	0.1 (0.09)
Challenging behaviours	-0.2 (0.61)	-0.02 (0.71)	-0.01 (0.84)	-0.01 (0.92)	0.01 (0.83)	0.01 (0.85)
Pharyngeal phase	0.1 (0.80)	0.4 (0.31)	0.3 (0.57)	0.6 (0.21)	0.3 (0.56)	0.4 (0.37)
Modified	0.1 (0.70)	0.6 (0.13)	0.4 (0.38)	1.0 (0.048)[e]	0.4 (0.38)	0.7 (0.16)
	Introduction of nutritional intervention					
	Supplementary feeding		**Gastrostomy feeding**			
	Crude OR (*P* value)	**Adjusted OR (*P* value)[c]**	**Crude OR (*P* value)**	**Adjusted OR (*P* value)[c]**		
OPD on DDS	2.6 (0.40)[d]	0.3 (1.0)	1.5 (0.76)[d]	n/c [d]		
Modified	8.1 (0.04)[d,e]	0.7 (1.0)	5.2 (0.13)[d]	n/c [d]		
OPD severity[a]	1.2 (0.02)[e]	1.0 (0.79)	2.1 (0.056)	n/c		
Feeding efficiency	1.1 (0.52)	1.2 (0.37)	0.9 (0.39)	0.8 (0.26)		
Challenging behaviours	1.0 (0.66)	1.0 (1.00)	1.0 (0.73)	0.9 (0.31)		
Pharyngeal phase	3.9 (0.23)	0.8 (0.88)	3.5 (0.26)[d]	0.8 (1.00)[d]		
Modified	7.3 (0.07)	1.4 (0.79)	6.3 (0.08)	0.8 (1.00)[d]		
	Parent Stress		**Hospitalisations for chest infection**			
	Crude OR (*P* value)	**Adjusted OR (*P* value)[b]**	**Crude OR (*P* value)**	**Adjusted OR (*P* value)[b]**		
OPD on DDS	1.1 (0.90)	0.5 (0.40)	1.5 (0.76)[d]	0.6 (1.0)[d]		
Modified	2.3 (0.14)	1.2 (0.83)	3.0 (0.34)	1.0 (0.98)		
OPD severity[a]	1.1 (0.02)[e]	1.0 (0.52)	1.1 (0.09)	1.1 (0.60)		
Feeding efficiency	0.9 (0.32)	0.9 (0.38)	1.0 (0.78)	1.1 (0.58)		
Challenging behaviours	1.1 (0.054)	1.1 (0.049)[e]	1.1 (0.54)	1.1 (0.59)		
Pharyngeal phase	2.8 (0.07)	2.0 (0.29)	2.0 (0.55)	0.8 (0.86)		
Modified	2.2 (0.12)	1.4 (0.58)	3.7 (0.26)	1.7 (0.67)		

Abbreviations: BMI, Body Mass Index; DDS, Dysphagia Disorders Survey; inf, infinity; n/c, not calculable; OPD, Oropharyngeal Dysphagia; PSAS, Pre-Speech Assessment Scale; SOMA, Schedule for Oral Motor Assessment

[a] Severity indicated by DDS raw score; [b]Model adjusted for GMFCS (collapsed I-II, III, IV-V, at 36 months), gender; [c]Model adjusted for GMFCS (at 36 months), BMI and gender; [d]Predicts perfectly, therefore calculated using Exact Logistic Regression; [e]Significantly related

Discussion

The classification and severity of OPD remained relatively stable between 18 to 24 months and 36 months, when removing classification error based on intra-child variability and limitations associated with typical development. The marginal reduction in OPD was seen as children with ambulatory CP (GMFCS I) matured. The modified OPD classification[2] accounts for the degree of maturation associated with typical development in the measure scores. Considering this, the change in OPD classification on the DDS may reflect later maturation of oral sensorimotor feeding skills in children with CP (particularly GMFCS I) compared to children with typical development.

The presence of an OPD classification did not change for children from GMFCS II-V following their second birthday, although OPD severity reduced in almost a third of children. The greatest and most frequent improvement in OPD severity was seen in children from GMFCS IV (on average 4.3 points). This may in part be due to small numbers in this group (n=4 with a DDS raw score), but may also relate to their heterogeneity in feeding skills.[32] Children from GMFCS IV also showed the greatest improvement of specific ingestion functions, which was particularly evident on pureed foods.

Children from GMFCS V appeared to reach their ceiling of performance for purees by 18 to 24 months (with all children impaired on all items, and no change between assessments). Interestingly more children from GMFCS V showed impairment on ingestion functions for chewable foods at 36 months compared to 18 to 24 months. This is perhaps due to the introduction of firmer chewable foods between these ages for children from GMFCS V, thus presenting more challenges. Similarly, more children from GMFCS III were impaired on fluid items *containment* (fluid loss) and *post-swallow* (coughing or wet respiration/ phonation) at 36 months. This may be explained by more children from this group using modified utensils at 18 to 24 months, but graduating to open cups or consecutive fluid swallows by 36 months. The developmental trajectories described in the gross motor literature,[33] suggest that children with poorer gross motor function will reach their functional capacity earlier than those with better gross motor function, which was reflected in our data. Gross motor function remained the best predictor of OPD classification and severity, being the only risk factor associated with each OPD outcome and at both assessment points (after adjustment for confounding).

During the 12 to 18 months between assessments, there were minimal changes in health outcomes. Regarding feeding method, only 1 child who was fed

orally (with modifications) at 18 to 24 months progressed onto tube feeding, and 1 who was predominately tube-fed transitioned to total tube-feeds. By 3 years, 9% of our sample received tube-feeding (a third of children from GMFCS IV-V), which was similar to average rates reported in a large multi-register study across 6 European countries (11%).[34] Regarding growth measures, on average children's weight- and BMI-for-age z scores reduced marginally by the second assessment, but height increased.

In order to facilitate earlier health management for children with CP, we were interested in understanding associations between OPD at 18 to 24 months and health outcomes at 36 months. The only health outcomes related to early OPD (accounting for GMFCS and gender) were weight, BMI, and parent stress. Weight and BMI z scores were related to the presence of OPD on the DDS (using modified cut-points). This supports the construct validity of the DDS as a measure which is detecting children at risk of later poor nutritional status. There were 27 children in our sample identified as having OPD on the DDS who were not underweight (BMI≤2SD), and 3 children without OPD who were underweight. Hence the DDS cannot be used in isolation from a comprehensive mealtime and nutritional assessment for indicating children at risk of poor growth. Children of parents who experienced stress during mealtimes demonstrated a significantly greater number of challenging behaviours during meals, but OPD on the DDS was not related to this outcome. This suggests a child's active resistance to mealtimes increases the likelihood of stressful mealtimes for parents, rather than the child's motor difficulty during ingestion.

This study is the first, to our knowledge, to explore changes to OPD prevalence and severity in transitional feeders with CP. It also contributed novel information regarding risk factors for OPD, and the relationship between early OPD and later health outcomes. This study had some limitations which may have influenced the interpretation of findings. The measurement of OPD using the DDS has been strengthened through conducting validation against children with typical development, and testing its reproducibility, in particular test-retest reliability. This provided information regarding the margin of error associated with repeated measures, as well as between-mealtime child variability in scores. While our findings were reported accounting for these differences, it is possible that the measurement error obscured some of the sensitivity of the DDS to detect change in feeding performance, and as such may represent a more conservative estimate of change.

Exploring OPD in 18 month-old children with CP restricted our sample size, as many participants only entered the study at 30 to 36 months, with CP diagnosis on

average only occurring at 13.3 months.[35] Many of the health outcomes of interest, such as gastrostomy feeding and hospitalisation for chest infection were present in only a small subset of the sample. While the strength of this study was our ability to explore relationships with a direct OPD measure, future register-based studies may strengthen our preliminary clinical findings in understanding risk between early OPD and later health outcomes.

Conclusion

The GMFCS remained a strong risk factor for OPD presence and severity. Raising awareness of this relationship for early intervention clinicians may assist in earlier screening and referral to feeding/ nutritional interventions. A more conservative monitoring approach should be taken for children classified as GMFCS I with apparent OPD before 2 years, as it appears many of these children's skills continue to mature up to 3 years. OPD classification remained consistent between 18 to 24 and 36 months for most children from GMFCS III-V. Many children from GMFCS III-IV showed improvements in OPD severity, suggesting this group may be prioritised for feeding interventions from as young as 18 months, even if OPD is mild. Children classified as GMFCS V tended to show minimal change after 18 to 24 months, and as such, approaches focusing on safety and nutritional efficiency should be prioritised. These findings may also facilitate more appropriately targeted nutritional and feeding interventions considering their influence on health. The presence of OPD at 18 to 24 months had the greatest influence on nutritional status at 36 months, but OPD severity did not. This suggests improving feeding skills alone may be insufficient to influence growth outcomes, and as such, interventions should holistically consider dietary intake in addition to oral sensorimotor skill development.

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Study ID: | | | | |

Qld CP Child: Growth, Nutrition and Physical Activity Feeding Questionnaire

Date of interview: | | | / | | | / | | | | | **Appt type (mths):** | |

Day / Month / Year

RA Name:

RA Signature:

Please complete this questionnaire and bring to your appointment.
This questionnaire will ask about various aspects of your child's eating and drinking ability, various aspects of his/her medical history that may affect your child and your feelings about your child's eating and drinking skills. Please answer the questions as best you can. If you are unsure of any questions, please ask the researcher when you attend your child's appointment. Thank you for being involved in our research!

1. Saliva Control

a. Please select the option which best reflects the frequency and severity of your child's drooling over the past week:

Frequency

- O 1. No drooling - dry
- O 2. Occasional drooling - not every day
- O 3. Frequent drooling - every day but not all day
- O 4. Constant drooling - always wet

Severity

- O 1. Dry - never drools
- O 2. Mild - only wet lips
- O 3. Moderate - wet on lips and chin
- O 4. Severe - clothes and objects get wet
- O 5. Profuse - clothing, hands and objects very wet

b. Is this frequency/severity of drooling typical for your child? O Yes O No

c. Is your child currently teething? O Yes O No

If you selected 1 for both Frequency and Severity, go to Question 3, otherwise please continue with Question 2.

2. Impact of Saliva

Please place an **X** on the line to indicate your estimation of your child's saliva control / drooling for the following questions.

a. How severe do you think you child's drooling problem is?

Not at all ——— Extremely severe | | . |

b. How often does your child have severe choking or coughing episodes due to saliva?

Never ——— Every day | | . |

c. To what extent does your child's drooling affect his or her life?

Not at all ——— Greatly | | . |

d. To what extent does your child's drooling affect you and your family member's life?

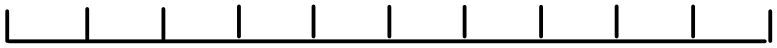
Not at all ——— Greatly | | . |

3. Types of food and fluids that your child eats and drinks:
Please indicate the types of food/fluids your child currently eats/ drinks:

Thin liquids [eg water, milk]	O Yes	O No
Nectar thickened liquids [Level 1 - mildly thick]	O Yes	O No
Honey thickened liquids [Level 2 - moderately thick]	O Yes	O No
Thickened liquids [Level 3 - extremely thick]	O Yes	O No
Puree (eg yoghurt)	O Yes	O No
Thick puree (eg smooth mashed potato)	O Yes	O No
Lumpy mashed foods (eg fork mashed vegetables, minced foods)	O Yes	O No
Chewable solids (eg bread, biscuits, fruits)	O Yes	O No
Tough chewable foods (eg meat not minced, lollies, and dried fruits)	O Yes	O No

4.Severity of eating or drinking problems:
Please rate on a scale of 1 to 10 whether, in your opinion, you think your child has any problems in eating and drinking compared to other children of his/her age.

Eating:

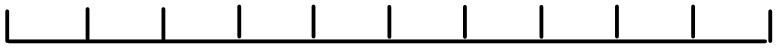


0 1 2 3 4 5 6 7 8 9 10

No problem Major problems

		.	

Drinking:



0 1 2 3 4 5 6 7 8 9 10

No problem Major problems

		.	

5. Presence of eating or drinking problems.
Which of the following statements best describes your child's feeding?

O 1. No feeding problems (eats normal diet and drinks for his/her age).

O 2. Mild swallowing or feeding difficulty (requires chopped/mashed foods).

O 3. Moderate swallowing or feeding difficulty and some difficulty with liquids (requires food well mashed/chopped and/or well moistened or requires liquids added to foods).

O 4. Severe difficulties with consuming liquids and foods.

O 5. Don't know.

6. Challenging behaviours demonstrated regularly during feeding:
Please indicate if your child demonstrates any of these behaviours at least daily during eating / meal-times:

Fussing or uncooperative during feeding	O Yes	O No
Fatigue/tiring/falling asleep	O Yes	O No
Distractability	O Yes	O No
Lack of interest	O Yes	O No
Overfilling/overstuffing mouth	O Yes	O No
Body stiffening or extending	O Yes	O No
Back-arching	O Yes	O No
Compressing lips to avoid food/fluids	O Yes	O No
Turning head away from food/fluids	O Yes	O No
Pulling or pusing food/fluids away	O Yes	O No
Throwing food or utensils	O Yes	O No
Batting the spoon/cup/plate	O Yes	O No
Vocal protesting or saying 'no'	O Yes	O No
Refusal	O Yes	O No
Crying/screaming	O Yes	O No
Spitting	O Yes	O No

7. Stress associated with feeding my child:

a. In general, the level of stress when trying to feed my child is …

O 1. Not at all stressful: the experience does not cause you to feel upset, tense or anxious
O 2. A little stressful
O 3. Moderately stressful
O 4. Very stressful
O 5. Extremely stressful: the experience upsets you and causes you a lot of anxiety or tension

b. I worry that my child does not eat enough food to grow properly…

O 1. Almost never / less than 10% of the time
O 2. A little bit / about 25% of the time
O 3. A moderate amount / about 50% of the time
O 4. Frequently / about 75% of the time
O 5. Almost always / over 90% of the time

8. Vomiting and Gastroesophageal Reflux:

a. Does your child vomit regularly? O Yes O No (Skip to Question 8e)

b. If your child vomits regularly, please indicate how frequently this happens: O Daily O A couple of times a week

c: If you child vomits daily, please indicate how many time this happens each day:

O Once a day O 2-5 times O 6-10 times O More than 10 times

d. Do you change your child's daily activities in any way to avoid/stop vomiting? O Yes O No

If yes, please describe (please print clearly)

e. Has your child ever been diagnosed by a doctor as having gastroesophageal reflux? O Yes O No

f. Does your child currently have a diagnosis of gastroesophageal reflux? O Yes O No

g. Does your child currently take medication for gastroesophageal reflux? O Yes O No

h. Has your child had a fundoplication for gastroesophageal reflux? O Yes O No

If yes, please describe (print clearly):

9. Food allergy or intolerance:

Does your child have an allergy or intolerance to any types of food? O Yes O No

If yes, please describe (print clearly):

10. Summary of my child's signs and symptoms during eating or drinking.
These are a list of signs and symptoms of swallowing difficulties that your child may demonstrate.
Please indicate if you see these signs/symptoms (at least once per day) during your child's eating or drinking and for which consistency/ies.

My child…….	Don't know	No	Yes	Thin Drinks	Thick Drinks	Smooth Puree	Lumpy Semi-Solid	Finger Foods
a. has difficulty drinking from a spout cup or cup.	O	O	O	O	O			
b. has difficulty moving the food to the back of his/her mouth when eating from a spoon.	O	O	O			O	O	
c. has difficulty biting and chewing his/her food	O	O	O				O	O
d. gags when eating or drinking.	O	O	O	O	O	O	O	O
e. coughs when eating or drinking.	O	O	O	O	O	O	O	O
f. chokes when eating or drinking.	O	O	O	O	O	O	O	O
g. vomits when eating or drinking.	O	O	O	O	O	O	O	O
h. clears his/her throat often during or after meals.	O	O	O	O	O	O	O	O
i. needs to swallow a number of times to clear each mouthful of food or drink.	O	O	O	O	O	O	O	O
j. wheezes during/after eating or drinking (Wheezing is a whistling sound from the chest during breathing).	O	O	O	O	O	O	O	O
k. has 'stridor' when breathing in or out during eating or drinking (Stridor is a harsh, high-pitched, vibratory noise in the throat particularly when breathing in.)	O	O	O	O	O	O	O	O
l. becomes breathless and breathes quickly during eating or drinking.	O	O	O	O	O	O	O	O
m. breathing becomes laboured or effortful during eating or drinking.	O	O	O	O	O	O	O	O
n. has a 'rattly chest' after eating or drinking.	O	O	O	O	O	O	O	O
o. gets a 'snuffly nose' after eating or drinking.	O	O	O	O	O	O	O	O
p. has a 'gurgly voice' after eating or drinking.	O	O	O	O	O	O	O	O
q. has runny eyes or 'eye tearing' after swallows of certain food or drinks.	O	O	O	O	O	O	O	O
r. seems to go 'blue' around the lips/face or turn 'dusky' or pale after drinking or eating.	O	O	O	O	O	O	O	O
s. generally refuses to eat or drink some food or fluid textures.	O	O	O	O	O	O	O	O

10. continued

t. Does your child regularly get high temperatures? O Yes O No

u. Has your child been diagnosed as having pneumonia in the last 6 months? O Yes O No

v. Has your child has any chest infections or respiratory conditions in the last 6 months? O Yes O No

If yes, please describe (print clearly)

11. Presence of tube feeding:

a. Does your child use a feeding tube at any time in the day or evening? O Yes O No (Skip to Question 12)

b. Please indicate which type of tube your child uses: O NG-Tube O G-Tube / G-J Tube / PEG

c. Please indicate the type of tube feeding regimen that your child has.

O Bolus feeds

O Continuous feeds

O Combination of bolus and continuous feeds

O Other

If other, please describe (please print clearly):

12. Method of feeding

Please indicate which of the following statements best reflects your child's feeding methods:

O 1. **Total oral intake:** All food and fluids by mouth with no modifications required

O 2. **Total oral intake with modifications:** All food and fluids by mouth, however food/fluids may be supplemented with extra calories or vitamins; fluids may be restricted or thickened; foods may be modified (e.g. pureed) or restricted (e.g. chewable foods); and/or special cups/spoons/utensils need to be used due to child's oromotor/swallowing skills.

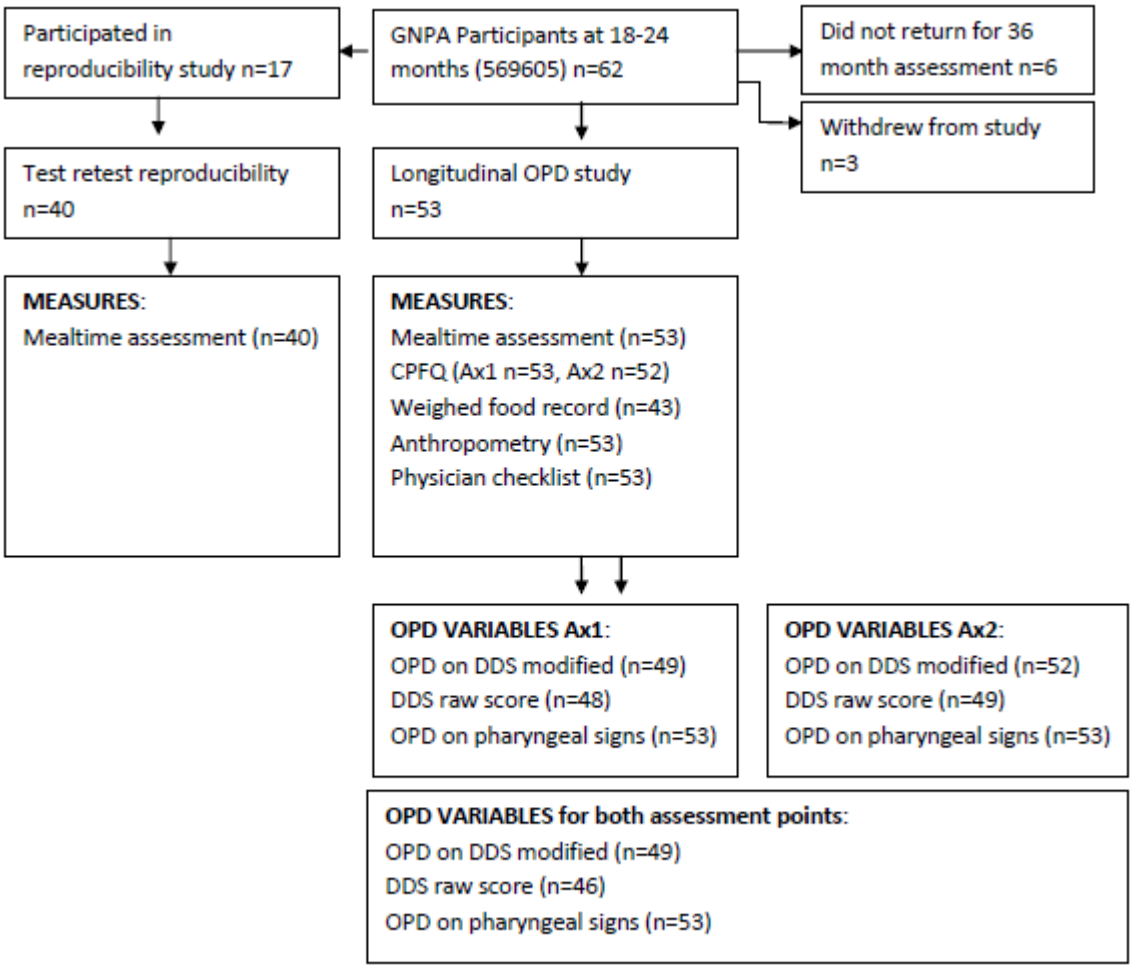
O 3. **Predominantly oral with supplemental tube feeds.** Most food is taken by mouth with small amounts of nutrition or hydration/fluids via the tube.

O 4. **Predominantly tube with small amounts of food or 'tastes' taken by mouth**. Most nutrition and hydration taken by tube, but allowed small amounts of oral foods (tastes) for pleasure or practice.

O 5. **Total tube feeding**: All nutrition and hydration via tube.

Thank you for completing the feeding questionnaire!
Please take this questionnaire along to your child's appointment with the researchers.

Supplementary Information 2. Flowchart of Recruitment Pathways



Abbreviations: CPFQ, Queensland Cerebral Palsy Child Feeding Questionnaire ; DDS, Dysphagia Disorders Survey; GNPA, Growth Nutrition and Physical Activity; OPD, Oropharyngeal Dysphagia; PSAS, Pre Speech Assessment Scale; SOMA, Schedule for Oral Motor Assessment; TD, Typically Developing

Supplementary Information 3. Changes to Oropharyngeal Dysphagia Proportion by Measure, Between 18 to 24 Month and 36 Month Assessments

	Test-retest %; kappa or ICC	18-24 months n (%) or mean (SD)	36 months n (%)	Change: n (%) or mean	Statistic (*P* value)
Overall OPD[b]	87.5; 0.2	47 (89)	42 (79)	11 (21)	2.3 (0.13)
SOMA (overall)	97.5; 0.95*	20 (38)	18 (34)	6 (11)	0.7 (0.41)
Puree	100.0; 1.0*	14 (32)	12 (26)	4 (10)	4.0 (0.046)
Semi-solid	100.0; 1.0*	8 (27)	9 (28)	0 (0)	n/c
Cracker	92.3; 0.8*	12 (27)	17 (35)	3 (7)	0.3 (0.56)
Bottle	n/c	7 (44)	8 (80)	2 (25)	0.0 (1.00)
Trainer cup	90.0; 0.8*	6 (21)	8 (40)	1 (8)	1.0 (0.32)
Cup	89.7; 0.6*	4 (21)	6 (16)	0 (0)	n/c
DDS-Part 2 (overall)	85.0; 0.3*	44 (83)	35 (66)	17 (32)	4.8 (0.03)
DDS-Part 2 (raw score)[a]	62.5; 0.9*	7.4 (7.3)	5.4 (7.3)	-2.0 (3.3)	-4.2 (<0.01)
Non-chewable score [a]	87.5; 0.9*	2.0 (2.5)	1.5 (2.5)	-0.5 (0.9)	-3.5 (<0.01)
Chewable score [a]	90.0; 0.8*	3.2 (2.8)	2.3 (2.8)	-0.8 (1.4)	-4.2 (<0.01)
Fluid score [a]	87.5; 0.8*	2.3 (2.5)	1.8 (2.4)	-0.6 (1.6)	-2.5 (0.02)
PSAS (overall)	87.5; 0.4*	33 (64)	38 (72)	13 (25)	1.9 (0.17)
PSAS Delay (binary)	87.5; 0.4*	31 (60)	38 (72)	15 (29)	3.3 (0.07)
PSAS Delay (score)[a]	66.7; 0.3*	15.5 (8.9)	17.4 (10.1)	1.9 (3.6)	3.1 (<0.01)
PSAS Disorder (binary)	92.3; 0.9*	26 (50)	20 (38)	12 (23)	3.0 (0.08)
PSAS Disorder (score)[a]	94.4; 0.8*	2.0 (3.2)	1.9 (3.2)	-0.1 (0.8)	-0.6 (0.54)
Pharyngeal signs overall	72.5; 0.3*	36 (68)	31 (59)	15 (28)	1.7 (0.20)
Gag	92.5; 0.5*	6 (12)	2 (4)	5 (10)	1.8 (0.18)
Cough	60.0; 0.2	25 (48)	10 (20)	22 (43)	8.9 (<0.01)
Wet breath	80.0; 0.5*	9 (17)	2 (4)	10 (20)	3.6 (0.06)
Gurgly voice	80.0; 0.5*	13 (25)	2 (4)	12 (23.5)	8.3 (<0.01)
Rattly chest	92.3; 0.7*	4 (9)	3 (7)	6 (17)	0.7 (0.41)
Choke	97.5; 0.7*	1 (2)	1 (2)	1 (2)	1.0 (0.32)
Vomit	n/c	0 (0)	0 (0)	0 (0)	n/c
Throat clear	80.0; 0.1	2 (4)	4 (8)	4 (8)	1.0 (0.32)
Multiple swallows	97.5; 0.9*	15 (29)	6 (12)	6 (12)	6.0 (0.01)
Wheeze	n/c	0 (0)	0 (0)	0 (0)	n/c
Stridor	n/c	0 (0)	0 (0)	0 (0)	n/c
Respiratory rate	92.5; 0.4*	2 (4)	2 (4)	4 (8)	0.0 (1.0)
Respiratory effort	92.5; 0.4*	3 (6)	0 (0)	2 (4)	2.0 (0.16)
Snuffly nose	92.5; 0.0	2 (4)	0 (0)	1 (2)	1.0 (0.32)
Eye tearing	95.0; 0.0	2 (4)	1 (2)	2 (4)	0.0 (1.0)
Circumoral cyanosis	90.0; 0.1	3 (6)	2 (4)	4 (8)	0.0 (1.0)

Abbreviations: GMFCS, Gross Motor Function Classification System; n/a, not applicable or available

[a]Agreement to +/- 1 point, and ICC**.**
[b] Overall OPD classification includes a positive case on the DDS, SOMA, PSAS or pharyngeal signs (unmodified).
*Indicates $P < .05$.

Summary of Chapter 9

The findings from this paper suggested that OPD is relatively stable between 18 to 24, and 36 months, although severity reduced between these ages. Early feeding was associated with poorer nutritional status and parent stress at 36 months. Specifically, it was found that:

i. Test-retest reproducibility for the DDS overall was strong, and for clinical signs was moderate. The OPD reclassification and change in OPD severity were described with reference to the smallest detectable change calculated in the reproducibility substudy (ie, a change in DDS scores that could be attributed to variability between children's mealtimes was not considered change for the longitudinal study).
ii. Overall, only 4 children who had had OPD at 18 to 24 months went on to be OPD-free at 36 months. Two children acquired an OPD classfication, with a net change of 2. Children from GMFCS I were the only group to show maturation of their OPD, suggesting the DDS was detecting children with delayed as well as disordered functions. OPD classification did not change for children from GMFCS II-V following their second birthday.
iii. About a third of children (30%) showed improvement in OPD severity, but did not necessarily have a change in OPD classification. Two children had worsening severity, 1 owing to the introduction of tube feeding. Improvement to OPD severity was noted across GMFCS levels.
iv. Children from GMFCS V appeared to reach their ceiling of performance for purees by 18 to 24 months, and performance on chewables worsened after this age.
v. Change in GMFCS between assessments was not related to change in OPD classification or severity. GMFCS appeared to be more strongly related to the presence of OPD at assessment 2, however more strongly related to OPD severity at assessment 1.
vi. GMFCS was the only risk factor associated consistently with OPD (presence and severity) between assessments. Age and epilepsy were also related to certain OPD outcomes but not consistently across OPD outcomes or assessment points.
vii. OPD on the DDS (modified) at 18 to 24 months was associated with low weight and BMI at 36 months. This was not significant using the unmodified DDS cut-points, which further strengthens the construct validity of our modification.
viii. The total number of challenging behaviours (at 18 to 24 months) was the only feeding variable associated with parent stress at 36 months.

ix. This study provided useful information to assist in prioritisation of which children to target for particular feeding interventions. A more conservative monitoring approach should be taken for children classified as GMFCS I with apparent OPD before 2 years. Children from GMFCS III-IV showed the greatest improvement, suggesting this group should be prioritised for feeding interventions from as young as 18 months, even if OPD is mild. Children from GMFCS V should have approaches focused on safety, and nutritional efficiency prioritised.

This chapter explored OPD longitudinally, and determined that GMFCS was the best predictor of OPD in preschool children with CP. The global burden of CP, however, is centred in low-resource countries, and as such an understanding of OPD in children with CP in this context is important. The following chapter sought to understand whether the findings from Australia, and other high-resource countries, could be generalised to a economically, geographically and ethnically contrasting country.

Chapter 10: Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy Studied in a High-Resource and Low-Resource Country

Introduction to Chapter 10

This chapter is comprised of the final substudy of this thesis, by way of a peer-reviewed paper “Motor Severity in Children with Cerebral Palsy Studied between a High-Resource and Low-Resource Country", and manuscript titled “Oropharyngeal Dysphagia in Children with Cerebral Palsy Studied in a High and Low Resource Country”. It has been estimated that 80% of the global burden of CP is in low-resource countries,[18] with little known about disability outcomes in these settings. Studying OPD in this often neglected majority is therefore of great importance.

The relationship between OPD and gross motor function and motor type has been well-established in the literature and throughout this thesis.The motor severity article, therefore, was written to provide a context for the OPD paper, to understand the gross motor functional severity and patterns in preschool children with CP, and how this differs from Australia. The OPD study aimed to compare the prevalence and severity of OPD in Bangladesh to the results of this doctoral study, thereby providing a greater understanding of the generalisability of the findings from Australia.

The Bangladesh fieldwork was conducted at the Centre for the Rehabilitation of the Paralysed (CRP), Dhaka district. This facility is a national rehabilitation centre providing services for families of children with CP from across the country. Bangladesh was selected as the location for this comparative study in a low-resource country as the doctoral candidate had previous relationships with the rehabilitation centre. The larger combined sample (of children from Bangladesh [n=81] and Australia [n=130]), allowed an additional analysis of the relationship between OPD and motor type/ distribution.

Paper 9: Motor Severity in Preschool Children with Cerebral Palsy Studied in a High-Resource and Low-Resource Country

This article was published in Pediatrics (journal impact factor 5.297). Reproduced with permission from Pediatrics, DOI:10.1542/peds.2014-1926.

Benfer KA, Jordan R, Bandaranayake S, Finn C, Ware RS, Boyd RN. Motor Severity in Children With Cerebral Palsy Studied in a High-Resource and Low-Resource Country. *Pediatrics.* 2014. http://pediatrics.aappublications.org/content/early/2014/11/18/peds.2014-1926.abstract. Accessed November 24, 2014.

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Motor Severity in Children With Cerebral Palsy Studied in a High-Resource and Low-Resource Country

Katherine A. Benfer, Rachel Jordan, Sasaka Bandaranayake, Christine Finn, Robert S. Ware and Roslyn N. Boyd

Pediatrics; originally published online November 24, 2014;
DOI: 10.1542/peds.2014-1926

Motor Severity in Children With Cerebral Palsy Studied in a High-Resource and Low-Resource Country

WHAT'S KNOWN ON THIS SUBJECT: There is variability in cerebral palsy prevalence estimates in low-resource countries, related to definitions, detection of milder cases, diagnosis age, and adequate training for clinicians. Thus, differences in prevalence and motor patterns between high- and low-resource countries remain unclear.

WHAT THIS STUDY ADDS: There were more children with dystonia and less with spasticity in Bangladesh compared with Australia (cerebral palsy diagnosis/motor classifications were consistent between settings). Differences in motor patterns between high- and low-resource countries have profound implications for early detection and appropriate interventions.

abstract

OBJECTIVES: To compare the patterns of motor type and gross motor functional severity in preschool-aged children with cerebral palsy (CP) in Bangladesh and Australia.

METHODS: We used comparison of 2 prospective studies. A total of 300 children with CP were aged 18 to 36 months, 219 Australian children (mean age, 26.6 months; 141 males) recruited through tertiary and community services, and 81 clinic-attendees born in Bangladesh (mean age, 27.5 months; 50 males). All children had diagnosis confirmed by an Australian physician, and birth and developmental history collected on the Physician Checklist. All children were classified by the same raters between countries using the Gross Motor Function Classification System (GMFCS), and motor type and distribution.

RESULTS: There were more children from GMFCS I–II in the Australian sample (GMFCS I, $P < .01$; III, $P < .01$; V, $P = .03$). The patterns of motor type also differed significantly with more spasticity and less dyskinetic types in the Australian sample (spasticity, $P < .01$; dystonia, $P < .01$; athetosis, $P < .01$). Birth risk factors were more common in the Bangladesh sample, with risk factors of low Apgar scores (Australia, $P < .01$), lethargy/seizures (Australia, $P = .01$), and term birth (Bangladesh, $P = .03$) associated with poorer gross motor function. Cognitive impairments were significantly more common in the Bangladesh children ($P < .01$), and visual impairments more common in Australia ($P < .01$).

CONCLUSIONS: Patterns of functional severity, motor type, comorbidities, etiology, and environmental risk factors differed markedly between settings. Our results contribute to understanding the patterns of CP in low-resource settings, and may assist in optimizing service delivery and prioritizing appropriate early interventions for children with CP in these settings. *Pediatrics* 2014;134:e1594–e1602

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KEY WORDS

gross motor severity, motor type, cerebral palsy, high-resource country, low-resource country

ABBREVIATIONS

CP—cerebral palsy
CRP—Centre for the Rehabilitation of the Paralysed
GMFCS—Gross Motor Function Classification System
OR—odds ratio

Ms Benfer designed the modifications to the Australian study for the Bangladesh context, collected the primary data in country, analyzed and interpreted the data, and drafted the manuscript; Ms Jordan completed the gross motor ratings, analyzed and interpreted the data, and drafted the manuscript; Dr Bandaranayake assisted in the modification of the protocol to the Bangladesh context, confirmed children's diagnosis and motor type, interpreted the data, and provided critical review of the manuscript; Ms Finn completed the gross motor ratings, interpreted the data, and provided critical review of the manuscript; Dr Ware advised on statistical design of the studies and the statistical analysis of the manuscript and provided critical review of the manuscript; Prof Boyd conceptualized the Australian study, assisted in the modification of the protocol to the Bangladesh context, secured funding for the Australian study, assisted in the interpretation of data, provided editorial support for the drafting of the manuscript, and provided study supervision; and all authors approved the final manuscript as submitted. All authors agree to be accountable for all aspects of the work to ensure its accuracy and integrity.

This trial has been registered with the ANZTR Register (identifier 1261200169820).

www.pediatrics.org/cgi/doi/10.1542/peds.2014-1926

doi:10.1542/peds.2014-1926

Accepted for publication Sep 8, 2014

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(Continued on last page)

Cerebral palsy (CP) is the most commonly occurring childhood physical disability,[1] with an overwhelming majority of its global burden in low-resource countries.[2] It has been estimated that 80% of the global prevalence of CP is in low-resource countries, having larger populations and potentially greater incidence rates.[2,3] Children who have a disability and their families living in low-resource countries are among the most disadvantaged in their community, with a bidirectional link between disability and poverty.[2] Bangladesh is a small but densely populated country in the Indian subcontinent (~150 million people, 150 000 km^2 land area). Almost a third live in extreme poverty (GDP per capita = US$ 752)[4] and ~45% of children aged <5 years have chronic malnutrition.[5] Australia, in direct contrast, is a large but sparsely populated country (22 million people, land mass of 7.7 million km^2)[6] and a major global economy (GDP per capita = US$ 67 442).[4,7]

Over the past decade, there have been a number of efforts to standardize the diagnosis of CP and motor type classification among western high-resource countries.[8–10] The prevalence of CP from various high-resource countries has been estimated at 2.0/1000 live births, and this has remained relatively stable throughout recent decades despite advances in medical practices.[11] Spasticity is typically cited as the predominant motor type, occurring in 77% to 93% of CP cases identified by a recent review, dyskinesia in 2% to 15%, and ataxia in 2% to 8%.[11] Of the 86.5% of individuals classified with spasticity in the Australian CP Register Report, 38.8% had hemiplegia, 37.5% diplegia, and 23.7% tri/quadriplegia.[12]

In low-resource countries there continues to be large variability in prevalence estimates, related to CP definitions, ability to detect milder cases, age at diagnosis (with CP prevalence influenced by survival rates), and adequate training for health staff.[3] Three population-based studies in low-resource countries have estimated prevalence as low as 1.6/1000 in urban China,[13] 2.8/1000 in India (4.4/1000 in children aged <4 years),[14] and as high as 4.0/1000 in Bangladesh (29.0/1000 in Dhaka district).[15] Studies of clinic attendees in these settings have reported high rates of spasticity similar to that in high-resource countries, from 70% to 90%.[14,16–20] Studies have tended to identify higher rates of quadriplegia than those reported in the west (60% to 86%),[16,17,20] although the only population-based study found a high rate of spastic diplegia (72.9%).[14]

Differences in prevalence and motor patterns between high- and low-resource countries reported in the literature remain unclear; however, the etiology has been reported to differ markedly. Owing to improved medical care in high-resource countries, it is now thought that birth asphyxia accounts for only 6% to 8% of CP cases,[1] with an increased proportion of preterm births (45%).[12] In low-resource countries there is poor survival of preterm infants, and home deliveries by unskilled birth attendants continue to dominate.[17] Birth asphyxia and low birth weight are reported as the prevailing causes of CP in low-resource countries,[16,17] along with kernicterus and postnatal causes such as meningitis and cerebral malaria.[3,14]

The motor outcomes of children in high-resource countries have been well described based on their level of gross motor function (Gross Motor Function Classification System [GMFCS]). Motor outcomes are impacted by a wide array of factors, including intrinsic child characteristics, family dynamics and functioning, and availability, access, and options for interventions.[21] Despite these many influences, gross motor functional development in western children who have CP has been shown to follow predictable patterns (along motor curves) based on the child's overall motor severity.[22] Less is known about the role of these environmental factors on motor outcomes in low-resource settings, where children may be in poverty, with less family knowledge and fewer resources to support their child's development, cultural differences in parental interaction style, and lower/delayed access to health services.[21,23]

Owing to these differences in neonatal risk factors and environmental influences between high- and low-resource countries, the severity and motor patterns of children who have CP in these 2 contexts is thought to differ. This study is the first to our knowledge to explore 2 cohorts of children who have CP in high- and low-resource settings using the same diagnostic and classification methods for each. It also aims to document differences in motor outcomes between settings with reference to gross motor function and motor type. This study will enhance our understanding of risk factors for CP and associated motor outcomes as well as contributing information to understand primary prevention priorities, and providing health ministries with data to plan optimal services.

METHODS

This article compares 2 cross-sectional prospective studies of children who have CP aged 18 to 36 months. The first sample is a cohort of children born in Queensland, Australia, and the second is a sample of clinic attendees residing in Bangladesh. The Australian data represent a subset of children from 2 larger longitudinal studies, Queensland CP Child Motor and Brain Development (National Health and Medical Research Council 465128)[24] and Queensland CP child: Growth, Nutrition and Physical Activity (National Health and Medical Research Council 569605).[25] It includes only initial assessments of children aged 18 to 36 months seen between January 1, 2009 and March 31, 2013.

Patients

Participants in Queensland were referred to the study through a range of settings from parent referral to community and tertiary care. All children who had a confirmed diagnosis of CP,[9] aged 18 to 36 months corrected age at initial assessment, and born in Queensland between 2006 and 2009, were invited to participate. Children who had neurodegenerative conditions were excluded.[24,25]

The Bangladesh sample was recruited through a national rehabilitation center in Bangladesh, the Centre for the Rehabilitation of the Paralysed (CRP). The center provides services to children who have CP residing in all regions of Bangladesh as outpatients, or through a 2-week inpatient program. The inpatient program provides parent education and training, as well as individual and group therapy. Admission is not associated with illness or medical intervention. All children aged 18 to 36 months who had a confirmed diagnosis of CP attending the center from August to December 2013 were invited to participate. Children who attended as inpatients were prioritized to enable a battery of measurements to be completed (for the larger study).

Procedures

For the Australian cohort, children attended the hospital for a diagnostic appointment with a pediatrician or child neurologist. During this appointment, diagnosis was confirmed based on published guidelines, and a detailed clinical history was taken. Children's motor type/distribution and GMFCS level were classified by 2 independent clinicians (pediatric rehabilitation physician, and an experienced physiotherapist).

In Bangladesh, children attended an initial diagnostic appointment with the primary investigator (KB) and a local pediatric physician, who collected the clinical history from the mother (in Bengali) and provided a preliminary diagnosis of CP. The written case history and a short video of the child performing functional motor tasks (including lying, rolling, sitting, standing, walking, and transitions between these) were sent to the Australian research team to provide an independent and consistent confirmation of CP diagnosis, motor type/distribution, and GMFCS.

Measures

The child's clinical history was collected by using the Physician Checklist (Supplemental Information A). This was administered by physicians to parents using open-ended questions to gather information on the child's clinical presentation, birth history, comorbidities, and development. This checklist was developed in 2003 for the Australian CP Child Study,[24] and was intended as a standardized physician checklist for gathering clinical history, rather than an exhaustive list of causes. Physicians made a judgment from the clinical history regarding factors potentially associated with a diagnosis of CP. Minor modifications were made to this checklist for Bangladesh (Supplemental Information B), which was translated from English into Bengali, and back-translated to confirm accuracy. Gestational age (time between the first day of the last menstrual period and child's date of birth) was recorded, and classified as term (>37 completed weeks of gestation), preterm (32 to <37 weeks), very preterm birth (28 to <32 weeks), and extremely preterm (<28 weeks).[26] The presence of comorbidities was collected from the parent in both contexts, however, using the standardized questions of the *10 Question Screen* in Bangladesh.[27] The socioeconomic status of Australian families was classified into tertiles using scores on the Socioeconomic Indexes for Areas Index of Relative Disadvantage.[28] The Poverty-Measurement Tool was used to classify the Bangladesh sample into 5 levels from well-off to poor, and has been validated in rural Bangladesh against an asset index and other traditional poverty measures.[29] The presumed timing (judged by physicians) of the complicating event was classified as antenatal, intrapartum, postpartum, or post-neonatal, or a combination of these (as reflected in Supplemental Information A). Five-minute Apgar scores <7 or a delayed cry >5 min after birth (in the absence of Apgars) were documented as a marker of neurologic depression.[17,30] Parents were also asked to report sitting and standing ability, and the age of acquisition of these skills.

Motor type/distribution were classified according to the Surveillance of Cerebral Palsy in Europe guidelines as spasticity (unilateral or bilateral), ataxia, dystonia, athetosis, or hypotonia.[10] The GMFCS classifies children into 5 levels, with the <2-year-old and 2- to 4-year-old scales used in the current study.[31]

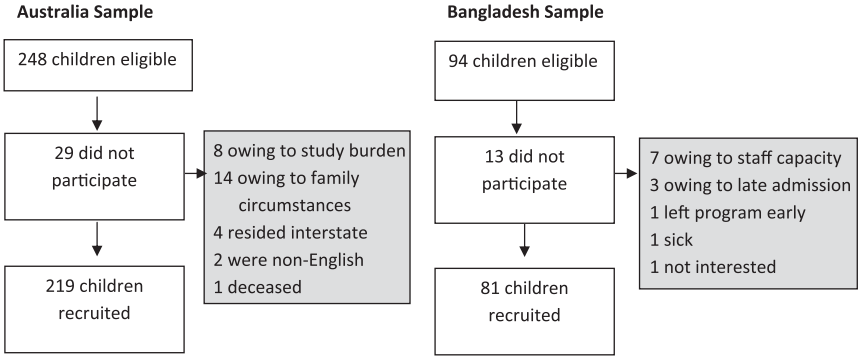

FIGURE 1
Recruitment pathways for Australia and Bangladesh samples.

Ethics

All families gave written informed consent to participate. The Australian study was approved by the Children's Health Services (Royal Children's Hospital Herston HREC07/QRCH/107), Southern Health Ethics (05077C), University of Queensland (2007001784), Cerebral Palsy League of Queensland (CPLQ2008/2009-1010), and Mater Health Services (1186C). Ethics for the Bangladesh Study were gained through the University of Queensland Medical Research Ethics Committee (2013000625), the Children's Health Services District Ethics Committee (HREC/13/QRCH/69), Centre for the Rehabilitation of the Paralyzed Ethics Committee (CRP/RE/0401/55), and the International Centre for Diarrheal Disease Research Bangladesh, Ethics Committee (PR-13047).

TABLE 1 Characteristics of Australian and Bangladesh Samples of Preschool-Aged Children Who Have CP

Sample Characteristic	Australia *n* (%)	Bangladesh *n* (%)	Crude OR (CI); *P* value (Bd base)	Adjusted OR (CI); *P* value
Gender			0.9 (0.5–1.5); .73	1.0 (0.6–1.7); .90
Male	140 (63.9)	50 (61.7)		
Female	79 (36.1)	31 (38.3)		
Preterm or term birth				
Extremely preterm	22 (10.0)	0 (0.0)	17.7 (2.1 to infinity); <.01[a]	NC
Very preterm	37 (16.9)	3 (3.9)	5.0 (1.5–16.7); .01	5.2 (1.5–18.0); .01
Preterm	35 (16.0)	15 (19.5)	0.8 (0.4–1.5); .48	0.6 (0.3–1.3); .22
Term	125 (57.1)	59 (76.6)	0.4 (0.2–0.7); <.01	0.5 (0.2–0.8); .01
Unknown	0 (0.0)	4 (4.9)		
Motor type				
Spasticity	192 (87.7)	50 (61.7)	4.4 (2.4–8.1); <.01	3.2 (1.7–6.0); <.01[b]
Unilateral	71 (32.4)	5 (6.2)	7.3 (2.8–18.8); <.01	3.5 (1.2–10.4); .03[c]
Bilateral (2 limbs)	49 (22.4)	21 (25.9)	0.8 (0.5–1.5); .52	0.4 (0.2–0.8); .01[d]
Bilateral (3 to 4 limbs)	72 (32.9)	24 (29.6)	1.2 (0.7–2.0); .59	2.8 (1.4–5.7); <.01
Dystonia	6 (2.7)	15 (18.5)	0.1 (0.1–0.3); <.01	0.2 (0.1–0.5); <.01[e]
Athetosis	3 (1.4)	7 (8.6)	0.2 (0.0–0.6); .01	0.2 (0.1–0.8); .03
Ataxia/hypotonia	18 (8.2)	9 (11.1)	0.7 (0.3–1.7); .44	0.8 (0.3–2.0); .62
GMFCS				
I	88 (40.2)	7 (8.6)	7.1 (3.1–16.0); <.01	7.7 (3.3–17.8); <.01[f]
II	37 (16.9)	12 (14.8)	1.2 (0.6–2.6); .70	1.1 (0.5–2.3); .81[f]
III	30 (13.7)	25 (30.9)	0.4 (0.2–0.7); <.01	0.3 (0.2–0.6); <.01
IV	28 (12.8)	14 (17.3)	0.7 (0.4–1.4); .31	0.7 (0.3–1.4); .30
V	36 (16.4)	23 (28.4)	0.5 (0.3–0.9); .02	0.5 (0.3–0.9); .03
Comorbidities				
Epilepsy	51 (23.4)	38 (46.9)	0.3 (0.2–0.6); <.01	0.7 (0.4–1.2); .12[g]
Vision	75 (34.2)	22 (27.2)	1.4 (0.8–2.5); .25	2.6 (1.4–5.1); <.01[h]
Hearing	16 (7.3)	2 (2.5)	3.1 (0.7–13.9); .14	4.4 (1.0–20.4); .06[i]
Speech	91 (41.6)	51 (63.0)	0.4 (0.3–0.7); <.01	0.6 (0.3–1.0); .06[j]
Cognitive	67 (30.6)	70 (82.7)	0.1 (0.1–0.2); <.01	0.1 (0.1–0.2); <.01[k]
Poverty status				
Well-off		25 (31.6)		
Moderately well-off		27 (34.2)		
Not so well-off		15 (19.0)		
Poor		7 (8.9)		
Very poor		5 (6.3)		
Unknown		2 (2.5)		
Socioeconomic status				
Least disadvantaged	73 (33.3)			
Middle tertile	54 (24.7)			
Most disadvantaged	92 (42.0)			

Adjusted OR models include covariates of GMFCS, age, gender, and preterm status, except when that variable is the main explanatory variable. Bd base, Bangladesh comparison group; CI, confidence interval; NA, not applicable to the context, therefore ORs not calculated; NC, not calculable, as no children in Bangladesh were extremely preterm.

[a] Calculated using episheet, Fisher's exact test.

[b] GMFCS significant (OR, 0.7, $P < .01$).

[c] Age (OR, 0.9, $P = .12$), GMFCS (OR, 0.3, $P < .01$), and preterm status (OR, 0.4, $P < .01$) significant.

[d] GMFCS (OR, 0.6, $P < .01$) and preterm status (OR, 3.1, $P < .01$) significant.

[e] GMFCS significant (OR, 2.6, $P < .01$).

[f] Age significantly related to GMFCS I (OR, 1.1, $P < .01$), GMFCS II (OR, 1.0, $P = .05$).

[g] GMFCS (OR, 2.0, $P < .01$) and preterm status (OR, 0.5, $P = .05$) significant.

[h] GMFCS significant (OR, 1.9, $P < .01$).

[i] GMFCS significant (OR, 1.7, $P < .01$).

[j] GMFCS (OR, 1.3, $P < .01$) and preterm status (OR, 0.6, $P = .02$) significant.

[k] Preterm status significant (OR, 0.5, $P = .03$).

Statistical Analysis

Data analyses were performed using Stata 10.0 (Stata Corp, College Station, TX; 2007), with significance at $P < .05$. Sample characteristics were presented descriptively. Differences between countries were compared by using logistic regression (odds ratios [ORs]) for binary outcomes and linear regression for continuous outcomes, using Bangladesh as the comparison group. Presence/absence of each motor type, GMFCS level, and extent of preterm birth were explored by using binomial regression. To account for differences in sample characteristics between Australia and Bangladesh, ORs were adjusted for age, gender, GMFCS level, and preterm status (except when that variable was the main explanatory variable) for the demographics; and age, gender, and GMFCS level for models exploring birth and environmental risk factors and motor outcomes. Multinominal logistic regression analysis was used to explore associations between etiologies and the outcomes of GMFCS and motor type.

RESULTS

A total of 342 children were referred to the studies, of which 300 participated, 219 in the Australian sample and 81 in the Bangladesh sample (recruitment pathways are shown in Fig 1). Children's ages ranged from 17 to 37 months, with equivalent mean ages between samples (Australia, 26.6 months, SD, 6.5; Bangladesh, 27.5 months, SD, 6.1; $P = .25$). The Australian sample was representative of a population-based sample with regards to gender ($P = .06$), GMFCS ($P = .09$), and motor type ($P = .53$).[12] There were significant differences in participant characteristics between Australia and Bangladesh, as shown in Table 1. Children from GMFCS III–V who had bilateral involvement had significantly higher odds of having visual impairment compared with children in GMFCS I–II who had unilateral/3-limb involvement in Australia (OR, 7.7, $P < .01$), but not Bangladesh (OR, 0.8, $P = .84$). The poverty status of the Bangladesh sample was not associated with GMFCS ($P = .92$) or motor type ($P = .58$).

The prevalence of birth risk factors (according to presumed timing) is presented in Table 2. Home births were more common in Bangladesh, occurring in 37 deliveries (45.6%), compared with only 4 (1.8%) in Australia. The majority of home births in Bangladesh (73.0%) were by an unskilled birth attendant, a further 21.6% by a nurse, and 5.4% by a family member. The influence of birth complications on motor severity and motor type is shown in Table 3.

Children's motor outcomes and associated environmental factors are shown in Table 4. On average, children from Bangladesh were diagnosed at age 27.5 months, despite mothers reporting concerns from age 8.8 months. Of the 23.5% of Bangladeshi children who had previous access to physiotherapy, all of their treatment was limited to passive stretching. In contrast, 92.2% of Australian children had previous access to physiotherapy, which used motor learning, functional therapy, neurodevelopmental therapy, postural management approaches, or a combination of these. Children from Bangladesh spent on average 71% of their day in passive positions (lying, sitting on mother's lap, being carried), and the amount of passive time was greater for children who had poorer gross motor function (GMFCS I–II, 46.1%; III, 52.0%; IV–V, 94.7%; r = 0.8, $P < .01$).

DISCUSSION

The patterns of functional gross motor severity, motor type, comorbidities, birth, and environmental risk factors all differed markedly between the high- and low-resource settings. Our Australian cohort is consistent with previous published data in high-resource countries, where mild CP (GMFCS I–II) constitutes 50% to 60% of any given population.[32] This pattern was skewed in the opposite direction in our Bangladesh sample, with only 23% of

TABLE 2 Prevalence of Birth Risk Factors for Australian and Bangladesh Samples of Preschool-Aged Children Who Have CP

	Australia *n* (%)	Bangladesh *n* (%)	Crude OR (CI); *P* value (Bd base)	Adjusted OR (CI); *P* value
Home delivery	4 (1.8)	37 (45.7)	0.02 (0.01–0.07); <.01	0.02 (0.0–0.07); <.01
Antenatal (only)	42 (19.2)	1 (1.2)	19.0 (2.6–140.3); <.01	19.5 (2.6–146.2); <.01
Intrapartum (only)[a]	34 (15.5)	3 (3.7)	4.8 (1.4–16.0); .01	5.5 (1.6–18.8); <.01
Postpartum (only)	9 (4.1)	49 (60.5)	0.03 (0.01–0.06); <.01	0.03 (0.01–0.06); <.01
Apgar <7/delayed cry	31 (14.1)	62 (76.5)	0.1 (0.0–0.1); <.01	0.1 (0.0–0.1); <.01[b]
Neonatal jaundice	7 (3.2)	19 (23.8)	0.1 (0.0–0.3); <.01	0.1 (0.0–0.3); <.01
Lethargy/seizures in 72 h	48 (26.7)	32 (41.6)	0.5 (0.3–0.9); .02	0.5 (0.3–0.9); .02
Antenatal plus intrapartum	47 (21.5)	2 (2.5)	10.8 (2.6–45.6); <.01	8.7 (2.0–37.4); <.01
Intrapartum plus postpartum	17 (7.8)	18 (8.2)	0.3 (0.1–0.6); <.01	0.3 (0.1–0.7); <.01
Antenatal plus postpartum	11 (5.0)	3 (3.7)	1.4 (0.4–5.1); .63	1.6 (0.4–6.1); .50
Antenatal, intrapartum, and postpartum complications	21 (9.6)	2 (2.5)	4.2 (1.0–18.3); .06	4.9 (1.1–21.8); .04
Hospital admission	123 (55.9)	51 (86.4)	0.2 (0.1–0.4); <.01	0.2 (0.1–0.4); <.01
Post-neonatal complications	20 (9.1)	6 (7.4)	1.3 (0.5–3.2); .65	1.1 (0.4–2.9); .86

Adjusted OR models include covariates of age, gender, GMFCS. Bd base, Bangladesh comparison group.

[a] Includes preterm birth.

[b] GMFCS (OR, 1.3, $P = .05$) and age (OR, 0.9, $P = .02$) significantly related on logistic regression.

TABLE 3 Association Between Birth Risk Factors and Outcomes of Motor Severity and Motor Type in Preschool-Aged Children Who Have CP in Australia and Bangladesh

Birth Risk Factors	Australia OR (CI); *P* value I–II, *n* = 125; III, *n* = 30; IV–V, *n* = 64	Bangladesh OR (CI); *P* value I–II, *n* = 19; III, *n* = 25; IV–V, *n* = 37
Association with motor severity		
Home delivery: GMFCS I–II (base)	Reference	Reference
III	NC[a]	1.0 (0.3–3.4); .97
IV–V	8.1 (0.9–73.7); .06	0.9 (0.3–2.6); .77
Antenatal: GMFCS I–II (base)	Reference	Reference
III	1.5 (0.7–3.5); .30	1.2 (0.2–7.7); .88
IV–V	1.2 (0.7–2.2); .59	0.8 (0.1–4.9); .76
Intrapartum: GMFCS I–II (base)	Reference	Reference
III	1.6 (0.7–3.6); .27	1.0 (0.3–3.7); .98
IV–V	1.2 (0.6–2.1); .66	0.9 (0.3–3.0); .89
Preterm: GMFCS I–II (base)	Reference	Reference
III	1.7 (0.8–3.8); .19	0.4 (0.1–1.6); .19
IV–V	0.8 (0.4–1.4); .41	0.2 (0.1–0.9); .03
Postpartum: GMFCS I–II (base)	Reference	Reference
III	3.1 (1.3–7.1); .01	2.8 (0.2–33.7); .41
IV–V	1.5 (0.9–3.5); .10	0.6 (0.1–3.3); .57
Apgar <7/delayed cry: GMFCS I–II (base)	Reference	Reference
III	3.4 (1.0–11.5); .05	0.6 (0.1–3.8); .60
IV–V	7.5 (3.0–18.9); <.01	0.2 (0.0–1.1); .06
Neonatal jaundice: GMFCS I–II (base)	Reference	Reference
III	2.9 (0.5–18.2); .26	0.5 (0.1–2.2); .35
IV–V	1.3 (0.2–8.2); .76	1.0 (0.3–3.4); .95
Lethargy/seizures in 72 h: GMFCS I–II (base)	Reference	Reference
III	1.0 (0.3–2.9); .97	1.0 (0.3–3.6); .97
IV–V	2.5 (1.2–5.2); .01	1.7 (0.5–5.7); .36
Hospital admission: GMFCS I–II (base)	Reference	NC[a]
III	1.9 (0.8–4.4); .13	
IV–V	1.5 (0.8–2.8); .18	
Postneonatal complications: GMFCS I–II (base)	Reference	NC[a]
III	0.7 (0.2–3.5); .71	
IV–V	1.3 (0.5–3.4); .66	
Association with motor type		
Home delivery: spasticity (base)	Reference	Reference
Dyskinetic	11.9 (1.0–145.0); .05	1.4 (0.5–3.8); .53
Ataxia/hypotonic	11.9 (1.6–90.0); .02	1.7 (0.4–7.2); .45
Antenatal: spasticity (base)	Reference	Reference
Dyskinetic	1.8 (0.4–7.3); .43	0.9 (0.2–5.0); .91
Ataxia/hypotonic	2.5 (0.9–7.1); .09	1.1 (0.1–10.9); .92
Intrapartum: spasticity (base)	Reference	Reference
Dyskinetic	1.7 (0.4–7.1); .45	1.0 (0.3–3.0); .95
Ataxia/hypotonic	1.2 (0.5–3.1); .72	3.2 (0.8–13.7); .12
Preterm: spasticity (base)	Reference	Reference
Dyskinetic	0.4 (0.1–1.8); .22	0.3 (0.1–1.1); .71
Ataxia/hypotonic	1.2 (0.5–3.0); .76	NC[b]
Postpartum: spasticity (base)	Reference	Reference
Dyskinetic	1.5 (0.4–6.2); .58	NC[b]
Ataxia/hypotonic	1.8 (0.7–4.7); .27	0.3 (0.1–1.4); .12
Apgar <7/delayed cry: spasticity (base)	Reference	Reference
Dyskinetic	3.2 (0.8–13.6); .12	0.7 (0.2–2.1); .50
Ataxia/hypotonic	1.2 (0.3–4.4); .79	0.5 (0.1–2.3); .38
Neonatal jaundice: spasticity (base)	Reference	Reference
Dyskinetic	NC[b]	1.2 (0.4–3.6); .80
Ataxia/hypotonic	1.9 (0.2–17.1); .55	0.4 (0.0–3.4); .39
Lethargy/seizures in 72 h: spasticity (base)	Reference	Reference
Dyskinetic	4.1 (0.9–19.3); .07	1.0 (0.4–2.9); .97
Ataxia/hypotonic	1.9 (0.6–6.3); .27	1.5 (0.3–6.6); .61
Hospital admission: spasticity (base)	Reference	Reference
Dyskinetic	0.6 (0.2–2.3); .45	2.6 (0.3–23.1); .41
Ataxia/hypotonic	0.8 (0.3–2.1); .70	0.7 (0.1–7.7); .79

children functioning at GMFCS I–II. Although spasticity was the dominant motor type in the Bangladesh sample, it was a lower proportion than that reported in previous studies in low-resource countries,[17] and significantly lower than the rates identified in our Australian sample. There was a significantly greater number of term births in Bangladesh, consistent with other studies from low-resource settings,[14,17,20] which would be expected in settings with poorer survival of children born preterm.[3] One explanation for the differences in motor severity and type could relate to our use of consistent raters and definitions across both settings, which gives greater certainty when comparing data. Furthermore, in a recent meta-analysis, spasticity was found to be significantly lower (~14%) in term-born children who had CP compared with those born preterm.[32] Higher rates of term births with asphyxia, severe jaundice, and post-neonatal complications have also been associated with quadriplegia and dystonia.[3] Low Apgar/delayed cry, lethargy/seizures, and term birth were all associated with poorer gross motor function in our study.

Epilepsy and speech and cognitive impairments were more common in the Bangladesh cohort, and visual and hearing impairments in the Australian cohort. Only visual and cognitive impairments were different once differences in GMFCS and preterm status between samples were accounted for, which were influencing the relationship. These patterns could reflect the sensitivity of the 10-Question Screen used in the Bangladesh sample, which has strong sensitivity to detect motor, cognitive, and seizure disorders, but lower sensitivity for vision and hearing.[27] This is particularly significant as universal screening of vision/hearing does not occur in Bangladesh.[27,33] The prevalence of epilepsy and speech impairments in our Bangladesh sample were comparable

TABLE 3 Continued

Birth Risk Factors	Australia OR (CI); *P* value I–II, *n* = 125; III, *n* = 30; IV–V, *n* = 64	Bangladesh OR (CI); *P* value I–II, *n* = 19; III, *n* = 25; IV–V, *n* = 37
Post-neonatal complications: spasticity (base)	Reference	Reference
Dyskinetic	1.3 (0.2–10.9); .82	0.8 (0.1–7.6); .81
Ataxia/hypotonic	1.2 (0.3–5.7); .81	4.5 (0.6–31.7); .13

[a] 0 value in base group, therefore not calculable.

[b] No children from outcome group had exposure of interest, therefore not calculable.

to previous work in a similar sample from Bangladesh; however, our estimates for visual and cognitive impairments were much higher, and lower for hearing impairments.[34] In the Australian sample, the presence of epilepsy and visual and hearing impairments was comparable to that reported in our national register report, with speech and cognitive impairments somewhat lower, perhaps owing to the younger age of our sample.[12]

Conducting research in a setting with low resources has unique challenges, particularly when aiming to provide a direct comparison with a high-resource setting. The most significant limitation to this study was the recruitment

TABLE 4 Prevalence of Environmental Factors and Motor Outcomes in the Australian and Bangladesh Samples of Preschool-Aged Children Who Have CP

	Australia	Bangladesh	Crude OR or β (CI); *P* value (Bd base)	Adjusted OR or β (CI); *P* value
Mean age of first concern, mo	NA	8.8	NA	NA
I–II		12.7		
III		8.8		
IV–V		6.8		
Mean age of diagnosis, mo	13.3	27.5	−14.2 (−16.4 to 12.0); <.01	−14.6 (−16.6 to −12.6); <.01[a]
I–II	14.5	26.4		
III	14.8	30.6		
IV–V	9.6	26.0		
Prior contact with physiotherapy, %	92.2	24.1	37.5 (18.4 to 76.7); <.01	102.7 (33.9 to 310.6); <.01[b]
I–II	88.8	5.6		
III	93.3	28.0		
IV–V	98.4	30.6		
Equipment: chair, %	34.2	6.2	7.9 (3.1 to 20.4); <.01	22.7 (7.8 to 65.8); <.01[c]
I–II	9.6	5.3		
III	46.7	8.0		
IV–V	76.6	5.4		
Equipment: mobility, %	17.8	1.2	17.3 (2.3 to 127.7); <.01	17.0 (2.3 to 128.4); <.01[d]
I–II	12.0	0.0		
III	46.7	4.0		
IV–V	15.6	0.0		
Able to sit, %	55.9	48.1	1.4 (0.8 to 2.3); .22	0.8 (0.4 to 1.5); .45[e]
I–II	69.8	84.2		
III	60.0	80.0		
IV–V	26.6	8.1		
Mean age of sitting, mo	12.5	14.9	−2.4 (−4.8 to −0.0); .05	−1.3 (−3.4 to 0.9); .24[f]
I–II	10.3	12.4		
III	17.9	17.0		
IV–V	18.4	14.7		
Able to walk, %	35.5	8.6	5.9 (2.6 to 13.3); <.01	3.5 (1.3 to 9.1); .01[g]
I–II	58.7	36.8		
III	6.7	0.0		
IV–V	3.1	0.0		
Mean age of walking, mo	22.5	20.9	1.6 (−9.3 to 12.4); .77	−1.1 (−7.1 to 4.9); .70[h]
I–II	20.1	20.9		
III	33.5	NA		
IV–V	NA[i]	NA		

Adjusted ORs include covariates of age, gender, and GMFCS (collapsed). Bd base, Bangladesh comparison group.

[a] Age (β = 0.7, *P* < .01) and GMFCS (β = −1.4, *P* = .01) significant.

[b] Age (OR = 0.9, *P* = .01), GMFCS (OR = 2.5, *P* < .01), and preterm status (OR = 2.8, *P* = .02) significant.

[c] GMFCS (OR = 5.2, *P* < .01) significant.

[d] Preterm status (OR = 3.2, *P* < .01) significant.

[e] GMFCS significant (OR = 0.3, *P* < .01).

[f] GMFCS (β = 4.4, *P* < .01) and preterm status (β = 2.9, *P* < .01) significant.

[g] GMFCS (OR = 0.1, *P* < .01) and age (OR = 1.1, *P* < .01) significant.

[h] GMFCS significant (β = 34.7, *P* < .01).

[i] No age recorded, but parent reported walking ability.

of a sample of clinic attendees in Bangladesh, which may limit generalizability to the population, although by adjusting the models for differences in gross motor function, we were still able to compare between samples. Over 80% of eligible children attending the center in Bangladesh were recruited to our study, with no systematic bias in their selection, although this recruitment rate was low compared with national prevalence rates. The sample was skewed toward rural families (being the predominant group accessing inpatient services at the center), and included more moderately well-off and well-off families than would be expected for the country.[29] Admission as an inpatient at CRP is not associated with illness or medical interventions, and as such is unlikely to skew the sample. There was no association between the poverty status of the Bangladesh sample and motor severity/type, which suggests economic factors were not biasing the motor patterns of those attending for services. Use of parent report for gathering much of the birth history may be biased by recall in both settings. This could be confounded further in Bangladesh, where a greater number of births are unregistered and occur at home.

CONCLUSIONS

This comparative study has implications for understanding the motor severity and patterns associated with CP. Differences in children's environment, both physical and opportunities provided in the home, may have an effect on children's motor outcomes and GMFCS level. Significantly fewer children from Bangladesh GMFCS IV–V were able to sit, and from GMFCS III able to walk, which may be reflected in their lower access to therapy and supportive equipment, as well as a large amount of time spent in passive activities. This raises questions regarding whether these children "catch up" to children of a similar level from Australia, or whether their poorer gross motor function is likely to persist. Studies using the Gross Motor Function Measure to assess specific gross motor tasks, and longitudinal studies to determine change across time, would help in understanding the applicability of motor curves and whether the prognostic aspect of the GMFCS is valid in this different cultural and economic setting.

This study provides useful information to assist with global perspectives on CP management. The high rates of term-born children who have CP in Bangladesh suggest scope for improved primary prevention, particularly through education and support of unskilled birth attendants.[3] The delayed age of diagnosis and access to appropriate treatments in Bangladesh represents an important window of opportunity for secondary prevention through early intervention. The findings from the current study suggest there is likely to be a significant subgroup of term-born children who have dystonia for whom early motor type diagnosis is more challenging. This group may also require access to different treatments, particularly the use of medications and careful consideration of the appropriateness of surgical interventions. Uptake of classification systems such as the GMFCS has been limited in Bangladesh, so improved training of health staff in such classification systems[35] as well as resources to support CP diagnosis and differential diagnosis of motor types would enable fast-tracked screening and appropriate, targeted interventions. Although there are many important factors to prioritize in low-resource countries, initiation of a centralized CP register, initially of clinic attendees, with consistent screening and definitions between centers, may assist in understanding the national picture of the diagnosis, and thereby better targeted management.

ACKNOWLEDGMENTS

We thank Ms Jannatul Ferdous (B Science, SpThy), Dr Sabera Bilkis (MBBS), Ms Hosneara Parveen (B Science Physio), Ms Sharmin Hasnat (B Science, SpThy), Ms Shoma (B Science Physio), and other staff at the Centre for the Rehabilitation of the Paralysed for their support with conducting the research in Bangladesh. We also acknowledge the support of International Centre for Diarrhoeal Disease Research (Dr Baitun Nahar and Dr Tahmeed Ahmed) in collaborating on this research.

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(Continued from first page)

PEDIATRICS (ISSN Numbers: Print, 0031-4005; Online, 1098-4275).

FINANCIAL DISCLOSURE: The authors have indicated they have no financial relationships relevant to this article to disclose.

FUNDING: Funded by the Australian National Health and Medical Research Council Postgraduate Medical and Dental Scholarship (1018264–KAB), Career Development Fellowship (APP1037220–RNB), and Project Grants (569605 and 465128). Funding was also received from The University of Queensland Graduate School International Travel Award (KAB).

POTENTIAL CONFLICT OF INTEREST: The authors have indicated they have no potential conflicts of interest to disclose.

Motor Severity in Children With Cerebral Palsy Studied in a High-Resource and Low-Resource Country

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Pediatrics; originally published online November 24, 2014;
DOI: 10.1542/peds.2014-1926

Supplemental Information

SUPPLEMENTAL INFORMATION A QUEENSLAND CEREBRAL PALSY CHILD – PHYSICIAN'S CHECKLIST (NATIONAL HEALTH AND MEDICAL RESEARCH COUNCIL 465128, BOYD ET AL 2013)

Diagnosis	
Cerebral palsy (definitions by Rosenbaum et al 2005, Badawi et al 1998)	*Yes/no/unsure*
Date diagnosed	
Age diagnosed	
Age at first assessment	*(by anyone)*
Mother's name at delivery	
Place of birth	
Gestation at birth	*Time between the first day of the last menstrual period and the date of birth*
Plurality	*M = miscarriage* *G = pregnancy* *P = delivery* *(eg, M^1 G^2 P^1)*
Birth order	
Birth weight	
Apgar scores	*x @ 1 min and x @ 5 min (if available)*
Ethnic groups	*Caucasian* *Asian* *African* *Hispanic* *Indigenous*

Etiology

Known Cause	
Timing of event	*Prenatal (1st/2nd/3rd trimester), perinatal, or postnatal (if known)*
Pregnancy complications/concerns/exposures	Antenatal
	Antepartum hemorrhage
	Pre-eclampsia
	Antenatal drug use
	Miscarriage/death of co-twin or triplet
	Maternal diabetes
	Pregnancy induced hypertension (weight gain, increased blood pressure, proteinuria)
	Intercurrent infection
	Past medical/surgical history of mother and fetus
	Trauma history
	Structural abnormalities in reproductive system (eg, incompetent cervix, bicorneal uterus)
	Exposures, occupational risk
	Assisted pregnancy
	Unexplained illness in pregnancy
	Bleeding in the third trimester
	Intrapartum
	Intrapartum fever (in mother)
	Preterm labor
	Meconium
	Breech
	Shoulder dystocia
	Postpartum (for neonate)
	Delayed cry (>5 min after birth)
	Neonate turned blue/needed oxygen
	Lethargy or seizures within 72 h of birth
	Neonatal jaundice (nonphysiologic, requiring treatment)
	Cord around neck
Post-neonatal cause (first 2 years of life)	CNS infection (eg, meningitis/encephalitis)
	Malaria
	Head injury
	Near drowning
	Tumor
	Trauma
	Cerebral malformation
	Epileptic seizures
Family pedigree (family history), insert diagram	
Example questions to ask family: any evidence of illness in the family on the maternal or paternal side; specifically any problems with development or intellect; presence of motor disorder, congential deformity, decreased motor function over time, in utero/death, disease	

Patterns of Motor Impairment

Motor type *(SCPE definitions, 2000)*	Spastic
	Ataxic
	Dyskinetic, dystonic
	Dyskinetic, choreoathetotic
	Hypotonic
Distribution	Bilateral/unilateral
	Number of limbs (based on activity or function, not passive testing of muscle tone) 1/2/3/4
Head circumference at birth, cm	
Head circumference, current, cm	

Functional Level

GMFCS level (Palisano et al 1997)	I/II/III/IV/V
Age at independent sitting (identified by parents)	
Age at independent standing (identified by parents)	
Age at independent walking (identified by parents)	
Upper limb/handedness	

Comorbidities

Epilepsy	No Yes (defined by 2 unprovoked seizures excluding febrile or neonatal seizures) If yes, still on medication: Yes/No
Seizure type and date of commencement	*Ask the family to describe exactly what they observed* *Generalized or partial* *Generalized: sudden onset of seizures that compromises responsiveness and affects the whole body* *Partial: seizures have focality, therefore symptoms reflect onset in 1 part of the brain*
Neonatal seizures	
Infantile spasms	*Infantile spasm, a specific type of spasm (symmetrical, axial)*
Controlled/not controlled	
Medications	*Medications for seizures or any other medications*
Visual impairment (after correction, on the better eye)	Normal Impaired Severely impaired (blind/no useful vision)
Hearing impairment (before correction, on the better ear)	Normal Impaired Severely impaired (hearing loss >70 dB)
Communication	*Expressive* *Receptive* *Both* *Unclassified*
Intellectual impairment	*SCPE classification* *Normal: IQ ≥85, attendance of regular school without support* *Borderline: IQ 70 to 84* *Mild impairment: IQ 50 to 69, basic literacy/numeracy* *Moderate impairment: IQ 20 to 49* *Severe impairment: IQ <20*

Other

Number of hospitalizations for chest infections in the past 6 mo (since last visit)
Number of episodes of pneumonia
Asthma
Number of episodes of asthmatic attacks in the past 6 mo (since last visit)
Cranial nerves
MRI *date and location*

SUPPLEMENTAL INFORMATION B ADDITIONAL QUESTIONS ON PHYSICIAN CHECKLIST FOR BANGLADESH SAMPLE 10 QUESTION SCREEN (DURKIN 1994):

1. Compared with other children, did your child have any serious delay in sitting, standing, or walking?
2. Compared with other children, does your child have difficulty seeing, either in the daytime or at night?
3. Does your child appear to have difficulty hearing?
4. When you tell your child to do something, does he/she seem to understand what you are saying?
5. Does your child have difficulty in walking or moving his/her arms or does he/she have weakness and/or stiffness in the arms or legs?
6. Does your child sometimes have fits, become rigid, or lose consciousness?
7. Does your child learn to do things like other children his/her age?
8. Does your child speak at all? (Can he/she make himself/herself understood in words; can he/she say any recognizable words?)
9. Can he/she name at least 1 object (eg, an animal, toy, cup, spoon)?
10. Compared with other children of his/her age, does your child appear in any way mentally backward, dull, or slow?

GASTROINTESTINAL HEALTH

Number of episodes of diarrhea in the past 12 months

- Three or more stools are passed in 24 hours that are sufficiently liquid to take the shape of the container in which they are placed

Type of diarrhea

- Acute watery diarrhea
- Persistent diarrhea, >14 days bloody diarrhea

Treatment of diarrhea used

Paper 10: Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy Studied in a High and Low Resource Country

This article is under review with Developmental Medicine and Child Neurology. It was also presented as a free paper at the 68th Annual Meeting of the American Academy of Cerebral Palsy and Developmental Medicine, September 2014, San Diego, United States.

Benfer K.A., Weir K.A., Bell K.L., Ware R.S., Davies P.S.W., Boyd R.N. Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy: Comparison between High- and Low-Resource Countries. *Dev. Med. Child Neurol.* 2014;56(Supp 5):78-79. (Abstract)

Oropharyngeal dysphagia in children with cerebral palsy studied in a high and low resource country

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What's Known on this Subject

Children with disabilities in low-resource countries have significantly compromised growth compared to their non-disabled peers. This has been linked to the child's inability to self-feed with little consideration of the influence of the child's oral sensorimotor skills.

What this Study Adds

Prevalence and severity of OPD were comparable between high- and low-resource countries once adjusted for differences in gross motor function. This supports the robust association between motor severity and OPD in children with CP, regardless of ethnicity and health resourcing.

Acknowledgements

We would like to thank Ms Jannatul Ferdous (B. Science, SpThy), Dr Sabera Bilkis (MBBS), Ms Hosneara Parveen (B. Science Physio), Ms Sharmin Hasnat (B. Science, SpThy), Ms Shoma (B. Science Physio), and other staff at the Centre for the Rehabilitation of the Paralysed for their support with conducting the research in Bangladesh. We would like to acknowledge the guidance provided by Dr Melanie Adams for the Bangladesh modification to the protocol. We would also like to acknowledge the support of icddr,b (Dr Tahmeed Ahmed) in collaborating on this research.

Abstract

Objectives: To determine the prevalence and severity of OPD in preschool children with CP in Bangladesh, and how this compares to Australian children. It was hypothesised that OPD would be more prevalent and severe in Bangladesh.

Methods: Cross sectional, comparison of two cohorts. 211 children with CP aged between 18-36 months, 81 in Bangladesh (mean age=27.6 months, 61.7% males), and 130 in Australia (mean age=27.4 months, 62.3% males). OPD prevalence and severity were analyzed using the Dysphagia Disorders Survey (DDS), clinical signs suggestive of pharyngeal phase impairment, Thomas-Stonell and Greenberg saliva severity scale and parent-report on a feeding questionnaire. Gross motor skills were classified using the Gross Motor Function Classification System (GMFCS), motor type and distribution.

Results: (i) Bangladesh (BD) sample: OPD prevalence=68.1%; OPD severity=10.4 (SD=7.9). Australia (AU) sample: OPD prevalence=55.7%; OPD severity=7.0 (SD=7.5). (ii) There was no difference in OPD prevalence or average severity between samples when stratified for GMFCS (OR=2.4, $P = .051$ and $\beta=1.2$, $P = .08$, respectively); OPD prevalence was associated with GMFCS in each sample (BD: OR=7.3, $P < .001$; AU: OR=2.6, $P < .001$).

Conclusions: Despite overall differences in the patterns of OPD between the Bangladesh and Australian samples, the prevalence and severity (when adjusted for the functional gross motor severity of the samples) was equivalent. This study provides support for the robust association between functional motor severity and OPD prevalence/ severity in young children with CP, regardless of ethnicity and health resourcing.

Introduction

Feeding difficulties or oropharyngeal dysphagia (OPD) are common in up to 85% of children with cerebral palsy (CP), and are directly linked to poor dietary intake and consequent undernutrition.[1,2] With the 2015 deadline of the Millennium Development Goals, the focus on undernutrition in under 5 year olds in low-resource countries is pertinent.[3] There is a paucity of research investigating OPD in young children with CP, much of which explores OPD in high-resource societies. Childhood disabilities are more prevalent in low-resource countries (LC), with an estimated 80% of the global burden of CP.[4]

Bangladesh is a densely populated country (approximately 150 million people residing in a country only 150,000km^2),[5] where one third of people live in extreme poverty, and chronic malnutrition persists in 45% of children under 5.[6] To our knowledge, only one

population-based household survey in Bangladesh has been conducted, which estimated prevalence of CP as 4 per 1000 live births.[7] In contrast to Bangladesh, Australia is a large sparsely populated country and considered a major global economy.[8] Our previous work described significant differences in the motor types, gross motor severity, and birth risk factors between these countries.[9] Compared to Australia, children attending services in Bangladesh were more frequently nonambulatory CP and more commonly dyskinetic motor types.[9]

Amongst the limited literature on feeding and nutritional status in children with disabilities in LC, there is consensus that children's growth is compromised compared to nondisabled peers.[10-16] This has been linked to the child's inability to self-feed with little consideration of the influence of oral sensorimotor skills. One intervention study in Bangladesh investigated OPD in children with CP,[13] but prevalence of OPD and risk factors were not reported. Three other studies have described OPD prevalence in LC, but have been limited to parent report or informal methods.[10,17,18] Two studies (Turkey and India) estimated the prevalence of OPD in children with disabilities in LC at approximately 70%.[10,17] A study in South Africa found 35% of children with CP had been referred for feeding assessment based on retrospective chart review (n=19).[18]

Consequently, this study aimed to determine the prevalence and patterns of OPD in children with CP in Bangladesh compared to Australia using direct objective assessments. It was hypothesised that OPD in Bangladesh would be more prevalent and severe compared to Australia when stratified for gross motor function. Gaining an understanding of these differences will assist disability and health organizations to better strategically target the provision of limited health resources.

Patients and Methods

This is a multi-site cross-sectional prospective study of preschool-aged children with CP; with a subset of samples from 2 larger studies in Australia[19,20] compared to clinic-attendees from Bangladesh. All families gave written informed consent to participate, with ethics approvals gained through the relevant institutional committees.[9,19-21]

Patients

Children with a confirmed diagnosis of CP[22] aged 18 to 36 months corrected age participated. Those with neurodegenerative conditions were excluded. The Australian sample invited all children born in Queensland from birth years 2006-2009. The Bangladesh sample was recruited through in-patient services at a national tertiary

rehabilitation facility, the Centre for the Rehabilitation of the Paralysed (CRP), from August-December 2013.

Procedures

Children in Australia attended the hospital for diagnosis and were followed-up for anthropometry, and video-taped gross motor and mealtime assessments. In Bangladesh, children and their families attended CRP for a 2-week carer training and therapy program. On admission, they attended an appointment with the primary investigator (KB) and local Paediatric Consultant, who provided a preliminary diagnosis of CP. Throughout the 2-week stay, children had mealtime and gross motor assessments videoed for later rating, and anthropometric measurements collected. The written case-history and gross motor video were sent to the Australian research team for confirmation of CP diagnosis, motor type/ distribution and Gross Motor Function Classification System (GMFCS). All gross motor ratings were conducted by the same 2 physiotherapists.

All mealtime assessments were conducted with children well positioned for 3 presentations of 4 textures (puree, lumpy, chewable and fluid)[21]. Children were then allowed to complete the snack as usual. Mealtime videos were later rated by the same paediatric speech pathologist (certified in the Dysphagia Disorders Survey, DDS) for both samples.

Measures

Measures of OPD were selected following comprehensive systematic review of the psychometric properties and clinical utility.[21,23] Three direct measures classified OPD; the DDS– Pediatric (Part 2) (DDS), clinical signs suggestive of pharyngeal phase impairment, and the Thomas-Stonell & Greenberg Saliva Scale. The DDS raw score was also used to indicate OPD severity. Parent report on the Cerebral Palsy Child Feeding Questionnaire (CPFQ) was an indirect measure of OPD classification and severity.

The DDS consists of binary judgments of eight ingestion functions of the oral, pharyngeal and oesophageal phase (maximum impairment score =22). Primary validation and reliability was conducted in adults with developmental disability (mean 33 years), and shown to be strong.[24,25] The paediatric version has been used and validated in children from 18 months.[26,27] The modified cut-points developed for children with CP aged 18 to 36 months were used in this study.[27]

A determination of pharyngeal phase impairment was noted if children demonstrated any one of 16 clinical signs, except a single cough on thin fluids.[28] Signs

included gagging, coughing, choking, vomiting, throat clearing, multiple swallows, wheezing, stridor, rapid or laboured breathing, wet breathing, gurgly voice, rattly chest, snuffly nose, eye tearing, or circumoral cyanosis/ duskiness.[28]

The Thomas-Stonell and Greenberg saliva scale is a semi-quantitative observational scale, indicating presence and severity of saliva loss on a 5-point ordinal scale (*no loss* to *profuse*).[29] Parents reported on their child's severity of eating problems and drinking problems using 2 visual analogue scales (*no problem* to *major problems*).

The GMFCS was classified using the <2 years and 2 to 4 year age-bands.[30] The type of CP (spasticity, dystonia, athetosis, hypotonia/ ataxia) and motor distribution (unilateral vs bilateral) were also classified.[31,32] Gestational age (time between first day of the last menstrual period and the child's date of birth) was classified as term (>37 completed weeks of gestation), preterm, very preterm birth, and extremely preterm.[33] The socio-economic status of Australian families was classified on the Socio-Economic Indexes for Areas (SEIFA) Index of Relative Disadvantage.[34] In Bangladesh, the validated Poverty-Measurement Tool classified families into 5 levels from well-off to poor.[35]

Children's nutritional status was determined by height, weight, and body mass index (BMI). Height/ length was measured using a portable stadiometer (Shorr Productions, Maryland USA), and segmental lengths used when a direct measure of height was not possible. Weight was measured to the nearest 100 grams using digital scales. Anthropometric data were converted to z scores according to World Health Organization reference data.[36]

Statistical Analysis

Characteristics of the Australian and Bangladesh samples were presented descriptively. Differences between samples were analysed using logistic regression for binary outcomes; linear regression for continuous outcomes; and ordinal outcomes (GMFCS, motor type, preterm level) analysed pairwise by level. Bangladesh was the comparison group, and models were adjusted for GMFCS, motor type, age, gender and preterm status (except when the covariate was the main explanatory variable). Differences between samples for OPD prevalence and severity were analysed using logistic (prevalence) and linear (severity) regression, with GMFCS and sample as interaction terms to account for potential between country differences. Risk factors for OPD were explored using multivariate regression. The association between OPD (presence and severity) and motor type was explored by combining the Australian and Bangladesh samples. All data analyses were performed using Stata 10.0 (Statacorp 2007).

Results

A total of 221 children with CP participated in this comparative study; 130 from Australia (AU) and 81 from Bangladesh ([BD], Supplementary Information 1 for recruitment pathways). The mean age in months (SD) was equivalent between samples (AU: 27.4 (5.3), BD: 27.5 (6.1), *P* = .80). Other sample characteristics differed significantly according to preterm status, motor type, GMFCS, and nutritional status (Table 1).

Prevalence and severity of OPD

The prevalence of OPD in Bangladesh was 68.1% compared to 55.7% in Australia. Once stratified for GMFCS, prevalence was equivalent between countries (OR=2.4, *P* = .051), with some exceptions between individual GMFCS levels (Figure 1). The prevalence of OPD increased with poorer gross motor function for both samples (AU: OR=2.6 (95% CI=1.8, 3.8), *P* < .001; BD: OR=7.3 (95% CI=2.8, 18.8), *P* < .001).

OPD severity based on the DDS and parent-report is shown in Figure 2. Mean DDS score (SD) differed between samples (AU: 7.0 (7.5), BD: 10.4 (7.9); β=-3.4, *P* < .001), but was nonsignificant after adjustment for differences in GMFCS distribution between samples (β=1.2, *P* = .08). Children's gross motor function was significantly related to OPD severity for both samples (AU: β=3.8, *P* < .001; BD: β=4.6, *P* < .001). OPD severity based on parent report was not significantly different between samples (AU: 2.9, SD=3.5, BD: 3.6, SD=3.5; β=-0.7, *P* = .14), but approached significance when adjusting for differences in GMFCS distribution (β=0.8, *P* = .051). Children's severity on specific textures differed between samples only for children in GMFCS V on non-chewables (mean score in BD 5.2 and 5.8 in AU, *P* = .02) and fluids (mean score in BD 4.8, and 5.7 in AU, *P* = .03).

Risk factors for oropharyngeal dysphagia

Prevalence and severity of OPD on the DDS differed between countries in the multivariate models (adjusted for gross motor and demographic factors, Table 2). Children had 1.9-3.5 times the odds of OPD with each increase in GMFCS, and significantly greater OPD severity. Preterm birth reduced children's likelihood of OPD based on the DDS, and resulted in lower OPD severity. Increasing age reduced the likelihood of OPD of the pharyngeal phase.

Presence and severity of OPD on the DDS was significantly greater for all motor types compared to children with unilateral spasticity, except for those with 2-limb bilateral spasticity and hypotonia (Table 3). A greater proportion of children with dyskinetic CP or 4-limb spasticity had clinical signs.

Table 1. Characteristics of Australian and Bangladesh Samples of Preschool Children with Cerebral Palsy

Sample Characteristic	Australia *n* (%)	Bangladesh *n* (%)	Crude OR (CI); *P* value (Bd base)	Adjusted OR (CI); *P* value[a]
Gender				
Male	81 (62.3)	50 (61.7)	1.0 (0.6, 1.8); 0.93	1.0 (0.5, 1.8); 0.90
Female	49 (37.7)	31 (38.3)		
Preterm or term birth				
Extremely preterm	19 (14.6)	0 (0.0)	19.3 (3.3, inf);<0.001[b]	NC[b]
Very pre-term	27 (20.8)	3 (3.9)	6.8 (2.0, 23.3); 0.002	7.3 (2.1, 25.9); 0.002
Preterm	20 (15.4)	15 (19.5)	0.8 (0.4, 1.7); 0.55	0.6 (0.3, 1.3); 0.18
Term	64 (49.2)	59 (76.6)	0.4 (0.2, 0.7); 0.001	0.4 (0.2, 0.8); 0.007
Motor type				
Spasticity	113 (86.9)	50 (61.7)	3.9 (2.0, 7.5); <0.001	2.8 (1.3, 5.8); 0.007
Unilateral	41 (31.5)	5 (6.2)	7.0 (2.6, 18.6); <0.001	2.6 (0.8, 8.5); 0.12
Bilateral (2 limbs)	30 (23.1)	21 (25.9)	0.9 (0.5, 1.6); 0.64	0.3 (0.1, 0.8); 0.009
Bilateral (3-4 limbs)	42 (32.3)	24 (29.6)	1.1 (0.6, 2.0); 0.77	2.9 (1.3, 6.4); 0.009
Dystonia	2 (1.5)	15 (18.5)	0.07 (0.02, 0.3); <0.001	0.1 (0.02, 0.5); 0.005
Athetosis	4 (6.2)	7 (8.6)	0.3 (0.1, 1.2); 0.09	0.6 (0.2, 2.6); 0.54
Ataxia/ hypotonia	11 (8.4)	9 (11.1)	0.7 (0.3, 1.7); 0.40	0.7 (0.3, 2.0); 0.55
GMFCS				
I	57 (44.2)	7 (8.6)	8.3 (3.5, 19.3); <0.001	8.2 (3.5, 19.6); <0.001
II	15 (11.6)	12 (14.8)	0.8 (0.3, 1.7); 0.49	0.8 (0.3, 1.8); 0.53
III	23 (17.8)	25 (30.9)	0.5, 0.3, 0.9); 0.028	0.4 (0.2, 0.8); 0.008
IV	12 (9.3)	14 (17.3)	0.5 (0.2, 1.1); 0.09	0.6 (0.2, 1.3); 0.17
V	23 (17.7)	23 (28.4)	0.5 (0.3, 1.0); 0.05	0.6 (0.3, 1.2); 0.12
Poverty status	NA		NA	NA
Well off		25 (31.6)		
Moderately well off		27 (34.2)		
Not so well off		15 (19.0)		
Poor		7 (8.9)		
Very poor		5 (6.3)		
Unknown		2 (2.5)		
Socio-economic status		NA	NA	NA
Least disadvantaged	48 (37.2)			
Middle tertile	40 (31.0)			
Most disadvantaged	41 (31.8)			
Tube fed	16 (12.3)	0 (0.0)	15.8 (2.6, inf); <0.001[b]	NC[b]
Nutritional status				
HAZ: mean (SD)	-0.9 (1.4)	-2.5 (1.4)	β=1.7 (<0.001)	β=1.5 (<0.001)
WAZ: mean (SD)	-0.3 (1.2)	-2.4 (1.4)	β =2.1 (<0.001)	β =1.8 (<0.001)
Underweight [c]	6 (4.6)	19 (23.5)	0.2 (0.1, 0.4); <0.001	0.3 (0.1, 0.8); 0.02

Abbreviations: Bd base, Bangladesh comparison group; CI, Confidence Interval; GMFCS, Gross Motor Function Classification System; HAZ ,Height for age z score; inf, infinity; NA, Not applicable to the context, therefore odds ratios not calculated; NC, Not calculable as no children in Bangladesh in outcome; OR, Odds Ratio; WAZ, Weight for age z score

[a] Adjusted Odds Ratio models include covariates of GMFCS, age, gender and preterm status, except when that variable is the main explanatory variable.
[b] Calculated using exact logistic regression as outcome predicts perfectly (adjusted OR not calculable).
[c]Based on BMI z score less than 2SD.

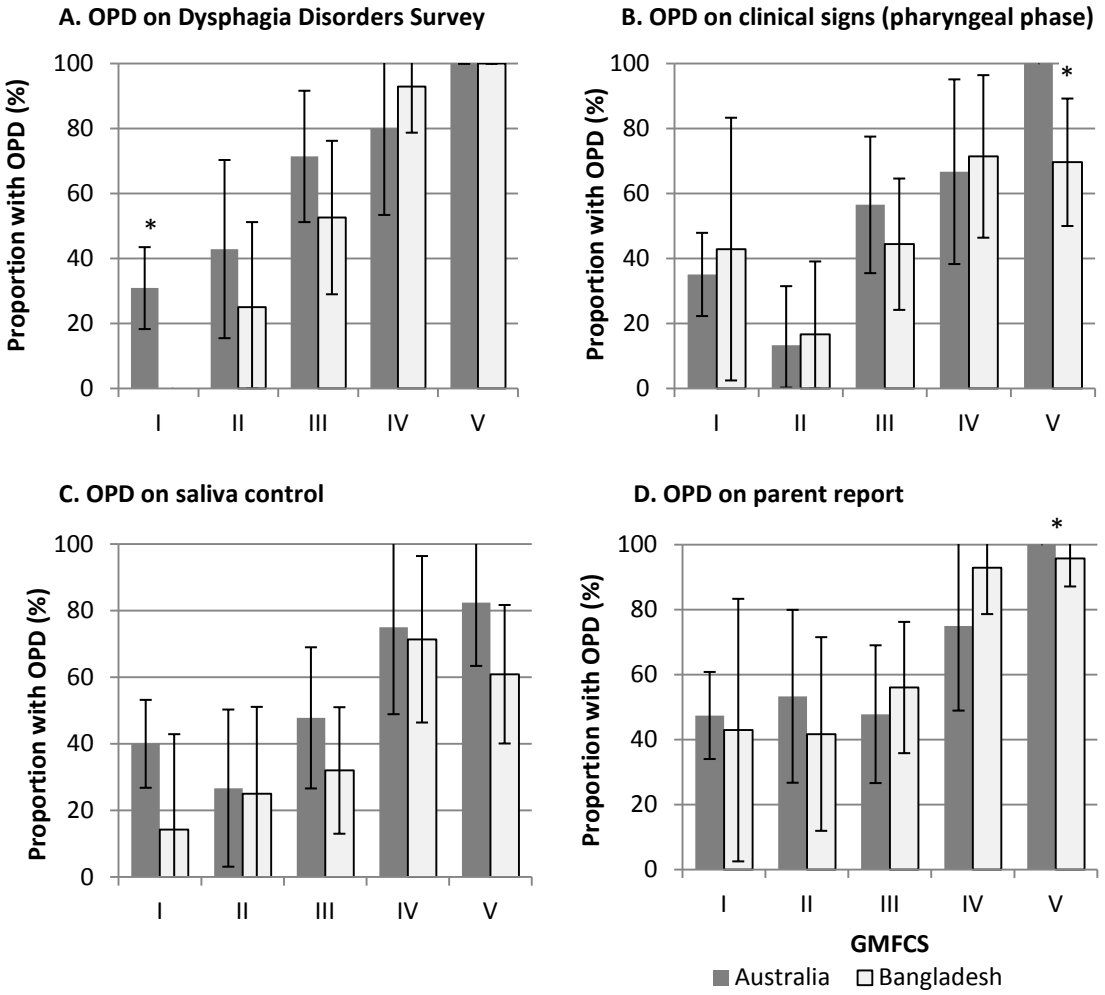
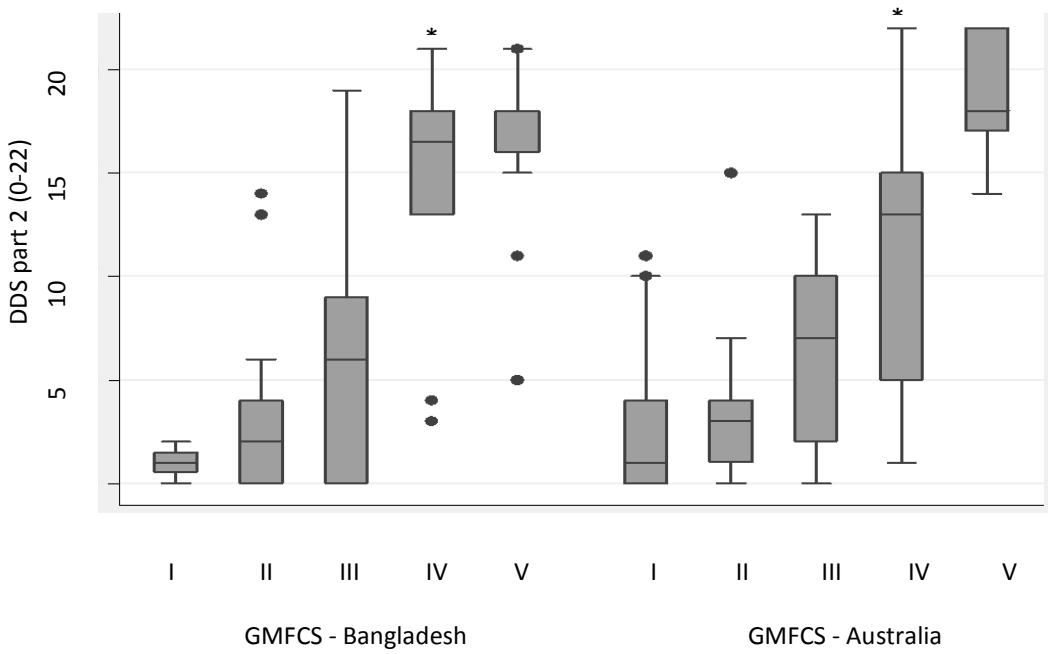


Figure 1. Prevalence of Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy in Australia and Bangladesh, According to Gross Motor Function

Abbreviations: GMFCS, Gross Motor Function Classification System; OPD, oropharyngeal dysphagia

*Indicates significant difference in proportion with OPD between samples; significantly more children in GMFCS I with OPD (DDS) in Australia compared to Bangladesh ($p<0.001$); significantly more children in GMFCS V with clinical signs suggestive of pharyngeal phase impairment in Australia compared to Bangladesh ($p<0.001$); significantly more children in GMFCS V with parent-reported OPD in Australia compared to Bangladesh

A. OPD severity on direct assessment (Dysphagia Disorders Survey)



B. OPD severity on parent report

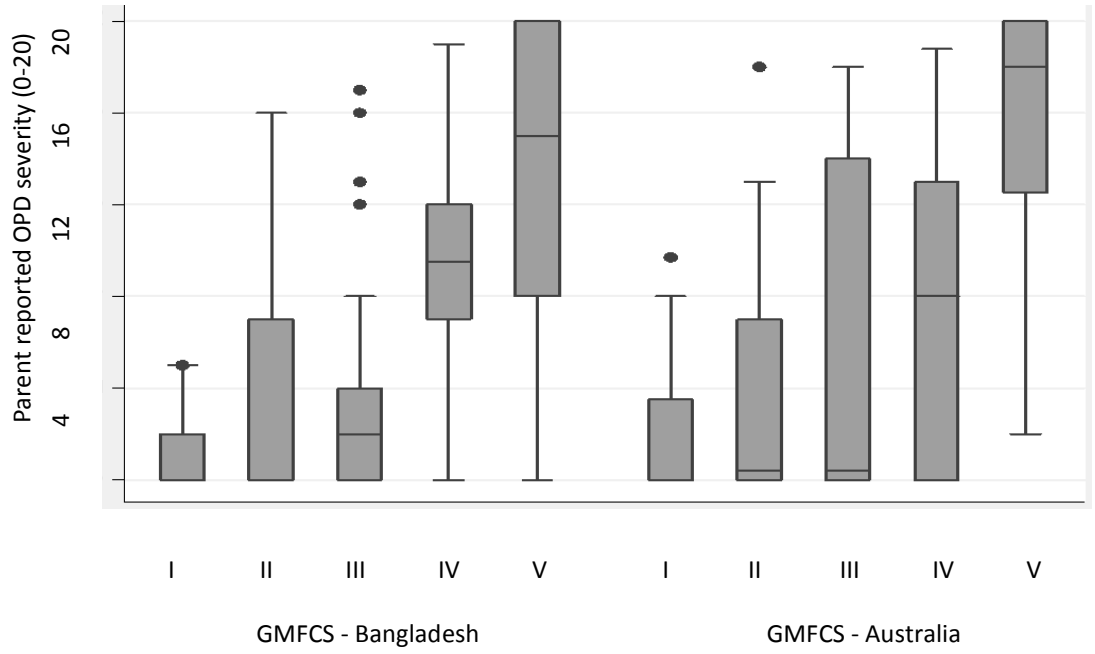


Figure 2. Severity of Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy from Australia and Bangladesh, According to Gross Motor Function: Direct Assessment and Parent Report

Abbreviations: DDS, Dysphagia Disorders Survey; GMFCS, Gross Motor Function Classification System; OPD, oropharyngeal dysphagia

Difference in mean DDS scores between samples by GMFCS level: I= 1.4 (-2.8, 5.5), *P* = .53; II= 0.4 (-2.8, 3.5), *P* = 83; III= 0.6 (-2.0, 3.2), *P* = .66, IV= -3.5 (-6.9, -0.2), *P* = .04; V= -2.3 (-0.1, 4.7), *P* = .060; Difference in mean parent-reported severity between samples by GMFCS level: I=0.4 (-1.6, 2.5), *P* = .69, II=0.0 (-2.0, 2.0), *P* = 1.0, III= -1.0 (-0.5, 2.4), *P* = .20; IV= -0.8, (-2.9, 1.3), *P* = .44; V= 1.2 (-0.3, 2.8), *P* = .10

Table 2. Risk Factors for the Presence and Severity of Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy

Sample Characteristic	**Adjusted statistic (CI); *P* value** (Bd base)
DDS (overall), OR	
Sample	6.9 (2.0, 24.3); 0.002
GMFCS	3.5 (2.2, 5.6); <0.001
Motor type	1.5 (1.0, 2.3); 0.06
Age	0.9 (0.9, 1.0); 0.07
Gender	0.9 (0.4, 2.1); 0.72
BMI	0.9 (0.6, 1.4); 0.69
Preterm	0.2 (0.1, 0.6); 0.002
Epilepsy	2.5 (0.7, 9.2); 0.17
DDS (severity), β	
Sample	2.0 (0.5, 3.6); 0.011
GMFCS	3.7 (3.2, 4.2); <0.001
Motor type	0.5 (0.0, 1.0); 0.06
Age	-0.2 (-0.3, -0.1); <0.001
Gender	0.3 (-1.1, 1.6); 0.71
BMI	0.1 (-0.5, 0.6); 0.80
Preterm	-2.3 (-3.7, -0.9); 0.001
Epilepsy	0.9 (-0.7, 2.5); 0.27
Pharyngeal phase, OR	
Sample	1.7 (0.8, 3.7); 0.21
GMFCS	1.9 (1.4, 2.5), <0.001
Motor type	1.0 (0.8, 1.3); 0.91
Age	0.9 (0.9, 1.0); 0.03
Gender	0.9 (0.5, 1.8); 0.80
BMI	1.1 (0.9, 1.4); 0.41
Preterm	0.9 (0.4, 1.7); 0.63
Epilepsy	1.3 (0.6, 3.0); 0.55

Abbreviations: BMI, Body Mass Index; CI, Confidence Interval; DDS, Dyspagia Disorders Survey; GMFCS, Gross Motor Function Classification System; OR, Odds Ratio

Discussion

Differences in Sample Characteristics Between Countries

The motor severity, motor type and other demographic characteristics of children differed significantly between Australia and Bangladesh.[9] Of particular importance is the distribution of GMFCS levels and motor type/ distribution, known to be related to OPD.[37] The Bangladesh sample was skewed towards children with poorer gross motor function (GMFCS III-V), whereas in Australia over half were classified as GMFCS I. In both samples, spasticity was the dominant motor type; however there were significantly fewer children with unilateral spasticity, and more children with dystonia in Bangladesh. When considering the generalizability of our findings to the CP population, it is important to note that participants in Bangladesh were all clinic attendees, and therefore may not represent the typical distribution of CP in Bangladesh.

Table 3. Motor Type and Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy

	OPD on DDS n(%)	**OR (95% CI); *P* value[a]**	**OPD on pharyngeal signs (%)**	**OR (95% CI); *P* value**	**Severity on DDS (mean, SD)**	**β (95% CI); *P* value**
Unilateral spasticity	13 (30.2)	ref	18 (39.1)	ref	2.5 (3.4)	ref
Bilateral spasticity (2)	16 (32.7)	1.1 (0.5, 2.7); 0.80	14 (26.9)	0.6 (0.2, 1.3); 0.20[c]	2.8 (3.8)	0.3 (-1.8, 2.4); 0.78[d]
Bilateral spasticity (4)	51 (86.4)	14.7 (5.5, 39.6); <0.001	45 (69.2)	3.5 (1.6, 7.7); 0.002	13.4 (7.1)	10.9 (8.9, 12.9); <0.001[e]
Hypotonia	3 (50.0)	2.3 (0.4, 13.0); 0.34	3 (42.9)	1.2 (0.2, 5.8); 0.85	4.8 (5.8)	2.3 (-2.4, 7.0); 0.34
Ataxia	9 (81.8)	10.4 (2.0, 54.9); 0.006	8 (61.5)	2.5 (0.7, 8.8); 0.16	9.7 (7.1)	7.2 (3.8, 10.6); <0.001
Athetosis	11 (100.0)	52.0 (4.9, inf); <0.001[b]	8 (72.7)	4.2 (1.0, 17.7); 0.055	16.6 (2.9)	14.1 (10.7, 17.5); <0.001[e]
Dystonia	14 (93.3)	32.3 (3.8, 272.0); 0.001	12 (70.6)	3.7 (1.1, 12.4); 0.031	15.4 (5.3)	12.9 (9.9, 15.9); <0.001

Abbreviations: DDS, Dysphagia Disorders Survey; inf, infinity; OPD, oropharyngeal dysphagia; OR Odds Ratio

[a] Relationship between motor type and OPD on DDS no longer significant once adjusted for GMFCS.
[b] Calculated using Episheet with continuity adjustment, as variable predicts outcome perfectly.
[c] Significantly lower proportion of children with bilateral spasticity (2 limb involvement) and pharyngeal signs compared to children with unilateral spasticity once adjusted for GMFCS (OR=0.3, *P* = .018).
[d] Significantly lower mean scores for children with bilateral spasticity compared to children with unilateral spasticity once adjusted for GMFCS (β= -2.4, *P* = .009).
[e] Mean score remained significantly higher compared to children with unilateral spasticity once adjusted for GMFCS

Most children with CP in Australia, however, access health services, making the samples comparable from a service-use perspective. Children with CP in Bangladesh had significantly lower height and weight z scores, and more were underweight compared to Australia, even after accounting for differences in GMFCS, preterm status, gender and age. This may in part be attributable to the child's OPD and its influence on dietary intake, but may also reflect the risk factors for high background rates of malnutrition in children in Bangladesh.[6] While inherent differences in body size related to ethnicity exist, use of the WHO classification for underweight status account for these.

Differences in Oropharyngeal Dysphagia Prevalence Between Countries

Presence and severity of OPD was greater in the Bangladesh sample compared to Australia, although more children in Australia used feeding tubes (absent in the Bangladesh sample). The prevalence estimate of OPD in Bangladesh was comparable to other LCs.[10,17] Due to variability in the samples and case ascertainment, comparisons are limited.

Differences in the sample recruitment may have influenced gross motor severity in these samples; as such, our a priori analysis plan was to stratify for GMFCS, accounting for some of these differences. Once stratified for GMFCS, differences in OPD presence and severity between countries were minimal, and in some instances lower in Bangladesh, contrary to our hypothesis. The only statistical differences were fewer children from GMFCS I having OPD (on the DDS) in Bangladesh, and fewer from GMFCS V having OPD (on the pharyngeal phase and parent report) in Bangladesh. This was surprising, considering children in Bangladesh have later diagnoses, and later and less access to therapy.[9]

A lower proportion of children from GMFCS V with clinical signs suggestive of pharyngeal dysphagia may be related to intrinsic and extrinsic factors. A much greater proportion of children from GMFCS V had dystonia in Bangladesh (30%) compared to only 4% in Australia. Also, mothers of children with nonambulatory CP in Bangladesh, were more likely to deliver very small amounts of fluids to their child and in a controlled mode of delivery (often from a teaspoon).

A trend for fewer children to have impaired saliva control in Bangladesh compared to Australia also existed (although nonsignificant). Children in Bangladesh had much lower fluid intakes, which may have resulted in dehydration and lower saliva production. Anecdotally, mothers in Bangladesh frequently wipe children's

saliva and food/ fluid loss during the mealtime, so the observations of saliva loss pre/post mealtime (as per the snack protocol) may not accurately reflect children's saliva control in Bangladesh.

Difference in Severity between countries:

Children in Bangladesh had more severe OPD, on average 3.4 DDS units (out of 22) higher than Australian children, although within the margin of smallest detectable change (based on reliability studies).[27] Again, once stratified for GMFCS, there were no differences in severity between countries, although there was an overall trend for less severe OPD by GMFCS level in Bangladesh. The exception was children from Australia classified as GMFCS IV having less severe OPD compared to Bangladesh. Australian children from GMFCS IV were the most heterogenous group regarding OPD status, with scores spanning the full range from 1 to 22 DDS units. OPD severity of children from GMFCS III in Bangladesh appeared more similar to the GMFCS IVs in Australia, suggesting specific motor aspects within the GMFCS are influencing their OPD. Similarly, in Bangladesh, it seemed that children from GMFCS IV were more comparable in their feeding skills to children in GMFCS V.

Another GMFCS level of interest in OPD severity was that of children from GMFCS I. In Australia, children from this level had more varied scores, whereas in Bangladesh they were closely clustered around the mean. There were only 7 children from GMFCS I in Bangladesh, predominately children with bilateral spasticity with 2-limb involvement. This is markedly different from the profile of children from GMFCS I in Australia with about 65% having unilateral spasticity. This may be a significant factor influencing the differences in OPD between countries, particularly on the presence of possible pharyngeal dysphagia (which was about half as common in bilateral 2-limb spasticity as for unilateral spasticity). Furthermore, in Australia, children were more independent feeders, and encouraged to bring their own foods (which corresponded to the standardised textures). In Bangladesh children tended to be fed by their mothers, and were provided with the standardised foods for the mealtime assessment. These factors possibly increased the complexity of the mealtime in Australia, particularly for children with mild OPD.

Risk factors:

The prevalence and severity of OPD increased markedly with each increase in GMFCS for Australia and Bangladesh. This was consistent with previous literature on this relationship.[26,37-42] Even after adjusting for other important health and demographic risk factors, GMFCS remained strongly related to OPD. Preterm status also reduced the likelihood of OPD and resulted in lower severity on the DDS, independent of its association with motor type/ severity. This supports reports in the literature of poorer gross motor functional outcomes associated with later neurological lesions.[4,43]

The presence and severity of OPD on the DDS was significantly greater for all motor types compared to children with unilateral spasticity, except for those with 2-limb bilateral spasticity and hypotonia. There were a greater proportion of children who had clinical signs for the dyskinetic CP and 4-limb spasticity subgroups. This analysis was made possible due to the higher rates of dyskinetic motor types in the Bangladesh sample, present in only very small numbers in western representative samples. Previous studies analysing the relationship between OPD and motor type have focused on number of limbs involved (ie, unilateral vs bilateral involvement) rather than the differences in movement patterns.[41,44] This is a significant contribution to our understanding of some of these minority motor types.

Limitations:

While the strength of this study its consistent raters and methods between countries, there were a number of limitations which may influence the interpretation of the findings. Most significantly was the recruitment of clinic-attendees in Bangladesh, as opposed to a population-based sample in Australia. This limits generalisability to the population; although analysis of the findings by GMFCS allowed comparisons to be drawn between country samples.

Due to recruitment of clinic-attendees in Bangladesh, only a small number of participants were classified as GMFCS I, as parents of children with minimal limitation to gross motor function appeared to be less likely to access services. Consequently wide confidence intervals were obtained for this subgroup in the OPD prevalence estimates, which should be interpreted with caution. The homogeneity of OPD severity in the GMFCS Is in Bangladesh, however, meant that the small numbers had minimal impact on this analysis.

While GMFCS-adjusted rates of OPD were lower in Bangladesh, the nutritional status of children told a different story. Of course, OPD is only 1 risk factor for poor nutritional status, and the influence of other risk factors for poor nutritional status operating across the paediatric population in Bangladesh warrant more detailed analysis. The DDS, however, consists of a series of binary judgments, not accounting for graded differences within each ingestion function. Possibly children in Bangladesh had more significant OPD, but the DDS lacked sensitivity to detect such differences.

Conclusions:

Despite overall differences in the patterns of OPD between the Bangladesh and Australian samples, the prevalence and severity (when adjusted for the functional gross motor severity of the samples) was equivalent. Children participating in the Bangladesh sample represent those commonly accessing services in Bangladesh, and as such mealtime and nutritional strategies (eg, feeding tubes) to support optimal health need greater implementation. This study provides support for the robust association between functional motor severity and OPD prevalence/ severity in young children with CP, regardless of ethnicity and health resourcing.

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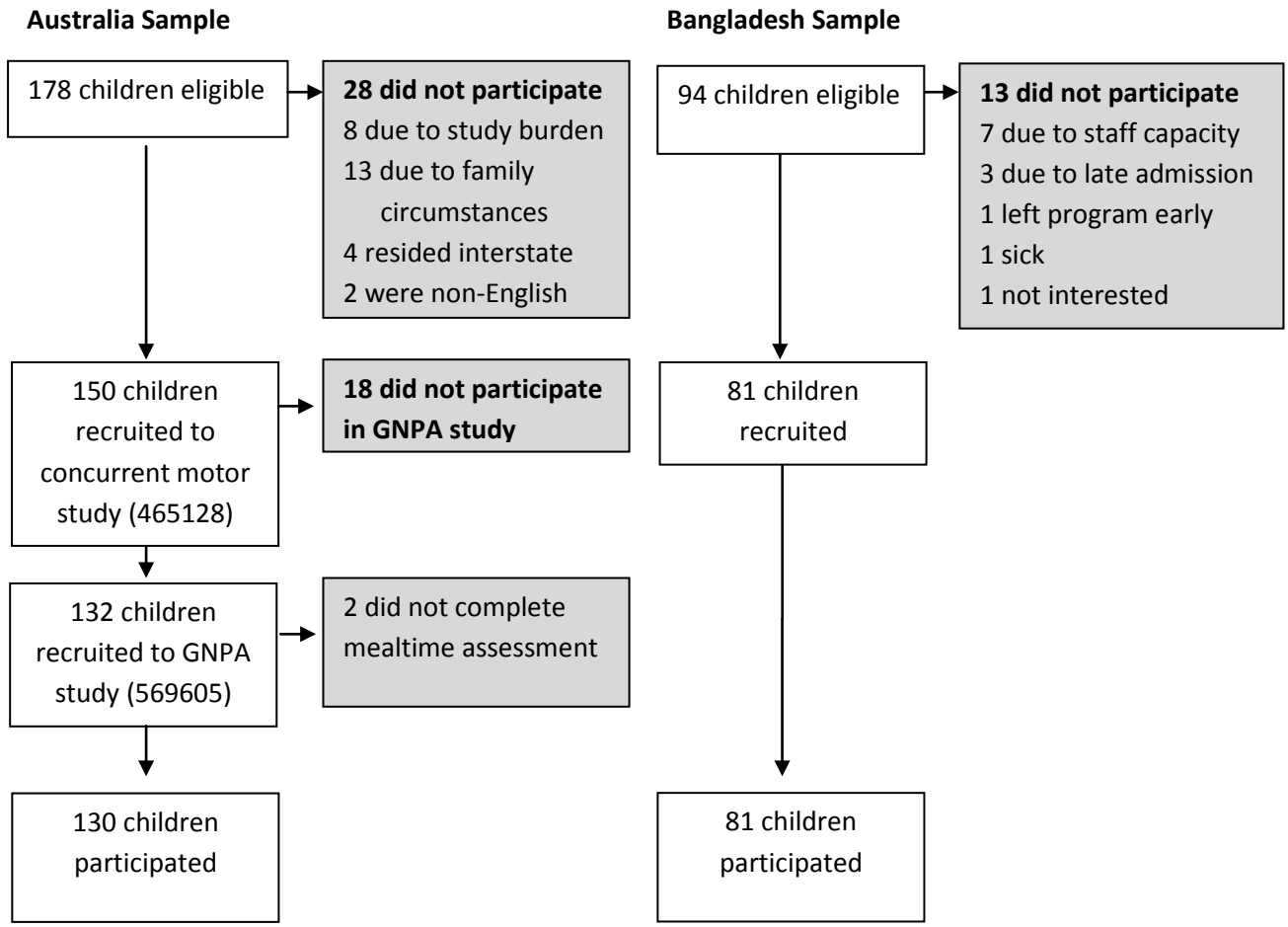
Supplementary Information 1. Recruitment Pathways for Australia and Bangladesh Samples



Key: GNPA Growth, Nutrition and Physical Activity (Study)

Summary of Chapter 10

These papers yielded some interesting findings regarding the consistency in the prevalence and severity of OPD between Australia and Bangladesh, despite overt cultural and economic differences between the 2 countries.

i. There were marked differences between the samples with regards to children's distribution of gross motor function (GMFCS) and motor type. Earlier work from this thesis has established that gross motor function and motor type are related strongly to OPD. The sample in Bangladesh was skewed towards children with poorer gross motor function (GMFCS III-V) whereas in Australia over half the sample was classified as GMFCS I. Bangladesh also had a more signficant subset of children with dystonia. Due to these differences found in our supplementary motor article, our a priori analysis plan was to stratify the sample by GMFCS to account for some of these differences, which would improve the interpretation of the OPD findings.
ii. OPD prevalence in Bangladesh in children aged 18 to 36 months (based on the DDS modified cut-points) was 68%, compared to 56% found in Australia. The average OPD severity was 10.4 in Bangladesh compared to 7.0 in Australia (out of a maximum score of 22).
iii. After stratification of the samples by GMFCS, there were no significant differences between Australia and Bangladesh for OPD prevalence or severity. Contrary to our hypothesis, there was a trend for fewer children in Bangladesh having OPD, and lower OPD severity. GMFCS was strongly related to OPD in both samples.
iv. The presence and severity of OPD on the DDS was significantly greater for all motor types (4-limb bilateral spasticity, dystonia, athetosis, ataxia) compared to children with unilateral spasticity, except for those with 2-limb bilateral spasticity and hypotonia. There were a larger proportion of children with clinical signs of pharyngeal phase impairment who had dyskinetic CP and 4-limb spasticity.
v. Our findings further supported GMFCS as a strong predictor of OPD prevalence and severity regardless of ethnic or economic factors.

The description of OPD in a low-resource country supported the generalisability of our findings to different populations. The final chapter of this thesis will conclude by summarising the findings from each of the substudies of this doctoral research.

Chapter 11: General Discussion and Conclusions

This chapter draws together the major findings from each of the 4 substudies, to provide an overview related to each of the study hypotheses. These findings are synthesised to present a clinical picture of the expected feeding, dietary intake and nutritional status of a preschool child with CP from each GMFCS level, and in comparison to the literature. This synthesis was included as part of the discussion, as it was considered the most clinically useful means of summarising the thesis findings and interpreting them in conjunction with the previous literature. The limitations of this doctoral research are discussed, as well as the implications of the findings for researchers and clinicians, and directions for future research in the field.

This thesis is the first to our knowledge to present data on OPD in preschool children with CP using direct standardised clinical assessments in a representative population-based sample. Most previous studies of OPD have been limited by using variable and often informal methods of data collection (including parent report). As such, the construct of OPD, and the psychometric properties of many of the measures of OPD have received limited attention, and there continues to be a lack of a gold standard in the field. A strength of the research described in this thesis was its in depth analysis of the measures, thereby better defining the construct of OPD. In order to achieve this, the first systematic review of OPD measures was conducted (Chapter 2), to report on the clinimetric properties of measures appropriate for use in preschool children with CP and neurodevelopmental disabilities. The psychometric properties of these measures were further explored through the validity and reproducibility study of the SOMA, DDS and PSAS (Chapter 5). This work used 2 new methods to contribute to understanding the properties of these measures; the inclusion of a typically developing reference sample, and use of latent-class variable analysis for *Tests in the Absence of a Gold Standard*.

The cross-sectional studies (Chapters 4, and 6 to 8) described in detail the OPD patterns that can be expected for preschool children with CP from each level of gross motor function, including those associated with the oral phase, pharyngeal phase and functional feeding on food/ fluid textures. Owing to the heterogeneity of a diagnosis of CP, and the changes associated with maturation of the oral sensorimotor mechanism and mealtime factors during childhood, this research sought to refine the sample to children aged 18 to 36 months, and describe impairments according to GMFCS. Including children with TD as a comparison group also allowed us to comment on OPD patterns present in children with ambulatory CP beyond those that are associated with typical development.

From these studies we found OPD was present among all levels of gross motor function, including those with ambulatory CP. Children from GMFCS I-II tended to have more isolated impairments, and those associated with feeding efficiency, whereas children from GMFCS III-V were more likely to have generalised impairments, and those associated with safety as well as efficiency. Better targeting OPD and nutritional interventions may be enabled by considering these patterns of OPD.

Two novel substudies (Chapters 8 and 9) contributed further to our understanding of the construct of OPD, and the influence of risk factors. OPD was explored in children longitudinally, between 18 to 24 and 36 months, to determine which children were likely to show maturation/ change in their OPD classification or severity by 3 years, and which ingestion functions were most likely to change. OPD was also explored in an ethnically and culturally contrasting country, Bangladesh, to look at differences in OPD in this low-resource context. Interestingly, in both of these studies, GMFCS was the risk factor which was most consistently associated with OPD presence and severity, despite age, ethnicity and health resourcing.

11.1 Overview

This overview will respond to the hypotheses proposed for the doctoral research. The hypotheses for each substudy will be addressed in turn.

Substudy 1: Validity and Reproducibility of Oropharyngeal Dysphagia Measures

H^{1A} The SOMA and DDS will be the most valid and reproducible direct clinical measures of OPD in young children with cerebral palsy. The PSAS will have the best clinical utility.

In order to determine the prevalence and patterns of OPD using direct standardised measures, a systematic review of the clinimetric properties of OPD measures was conducted. This review, presented in Chapter 2, found 9 measures of OPD which met inclusion criteria from a total of 27 papers. The COSMIN Checklist was used to objectively evaluate the psychometrics of measures, and has demonstrated validity and reliability.[79,95] This article found that the SOMA and FFAm were the 2 measures with the best published validity and reproducibility. These 2 measures (and the Gisel Video Assessment) had a much greater number of published papers testing their psychometric properties (owing to more frequent use in research), which would have influenced these findings. Clinical utility of the measures was evaluated using the Can Child Outcome Rating Form, and measures

were scored based on the measure's manual, clarity and the detail of information it yields. The SOMA and DDS had the best clinical utility, largely owing to their continued publication and the availability of manuals. While the PSAS did not score as well for its clinical utility (due to its lengthy and out-dated manual), it was acknowledged that it provides comprehensive information to support clinical decision making.

H^{1B} The prevalence of OPD detected on the PSAS will be equivalent to that on the DDS, but greater than that on the SOMA.

On initial evaluation, the PSAS, DDS and SOMA all appear to be measuring a similar construct of OPD. All 3 are focused on the oral phase, but each have a small number of items pertaining to the pharyngeal phase. All 3 measures cover the same three food/ fluid textures (puree, chewable and fluid), although with the addition of semi-solid foods in the SOMA. The method of measure development, scoring structure, and specific items tested in all 3 differ markedly, and as such it was expected that there would be variability in the prevalence detected by each. There have been no studies to date which have compared the prevalence of OPD detected on standardised measures. The findings of the validity and reproducibility study supported this hypothesis, with roughly equivalent proportions of OPD detected in children using the PSAS (73%) and DDS (85%), and only 35% on the SOMA. The PSAS and DDS had 85% agreement, suggesting they are in fact measuring a similar construct.

H^{1C} The specificity will be highest for the SOMA, but lower for the DDS and PSAS.

The diagnostic accuracy of measures (sensitivity and specificity) is important to know in order to interpret the measure results. This is typically calculated by comparing the diagnostic test result to the 'true' presence of the outcome, usually based on a gold standard measure. In the context of OPD, no gold standard test exists. The specificity of OPD measures was therefore calculated in this research using 2 methods. A sample of 40 children with TD was recruited, with the assumption that this sample was OPD free. Using this method, the specificity of the DDS was only 50%, the SOMA 100% and PSAS 63%. The second method used latent class variable analysis for *Tests in the Absence of Gold Standards*.[96] This web-based calculation yielded specificity results which were approximately equivalent to our calculation using the TD sample, of 47% for the DDS, 100% for the SOMA and 71% for the PSAS. These findings support the hypothesis

showing the specificity of the SOMA to be highest, and it to be lower for the DDS and PSAS. The latent-class analysis also provided results for the sensitivity of measures (DDS=100%, SOMA=53%, PSAS=100%), however future work using an expert speech pathologist panel as the gold standard would be valuable to confirm these findings.

H^{1D} There will be excellent reproducibility (test-retest, intrarater, interrater) for all OPD measures (including overall, the SOMA, DDS, PSAS, clinical signs).

Reproducibility describes the extent to which a measure can give repeatable results. It includes both agreement (how close the results are between ratings) and reliability (how distinguishable the results are between ratings). Interrater agreement was >85% for all binary published measures (SOMA, DDS, and PSAS), intrarater agreement was >90% and test-retest agreement >85%. Clinical signs overall were in agreement 90% for interrater, 95% for intrarater and 73% between mealtimes (test-retest). These findings support the hypothesis that reproducibility would be strong.

H^{1E} Parents will detect clinically significant/ overt OPD but will underdetect mild OPD.

Parents have been reported in the literature to underdetect OPD in their child with CP compared to direct assessment.[1] Our study found there was only moderate agreement (40% to 60%) between all parent reports and direct OPD assessment. This was true for the severity of oral phase impairments (48% to 58% for solids, and 39% to 63% for fluids), clinical signs suggestive of pharyngeal phase impairments (60%) and parent-reported ability on food/ fluid textures (40% to 60% depending on the measure). Interestingly, parents did not consistently overestimate or underestimate children's feeding severity, or the number of clinical signs (almost no bias for both, mean of differences<1.0). Agreement was better when considering the child's swallowing safety on textures (70% to 80%), which supports the hypothesis that parents may be better at detecting more clinically overt OPD. It may also be that parents do not consider specific oral phase impairments (such as limitations to biting, or drinking from a cup) as *limited ability* on that texture (which was the question parents were asked on the *Pediatric Evaluation of Disability Inventory*), but that they are accurately identifying many food/ fluid textures for which their child may need referral or have excluded from their diet. Training parents to detect safety concerns on food/ fluid textures may be more clinically meaningful and effective than focusing on their identification of specific oromotor impairments.

11.1.1. Summary of Oropharyngeal Dysphagia Measures for Preschool Children with Cerebral Palsy

Selecting the most appropriate measure for detecting OPD in preschool children with CP is critical for future research and optimal service planning. All 3 measures had strong agreement between raters for detecting OPD, and so 1 measure should not be prioritised for use based on this property. The PSAS and DDS were in agreement regarding case status about 85% of the time, which suggests they are both capturing a similar construct of OPD. Both the present study and previous literature have suggested the SOMA is detecting clinically significant OPD, but it lacks sensitivity to detect milder cases. It could be argued that this clinically significant OPD is the construct with which we should be concerned, with regards to potential impact on health outcomes. The association between various constructs of OPD and health outcomes, however, have not been well demonstrated in the literature (particularly as assessed by standardised measures). Our longitudinal study found that OPD on the DDS using modified cut-points at 18 to 24 months was significantly related to low weight and BMI at 36 months. Other standardised measures were not related to health outcomes in the current study.

As such we advocate use of a measure with greater sensitivity (ie, the DDS or PSAS) to ensure we are detecting the extent of the impairment. When using 1 of these more sensitive measures, we do not want to grossly overdetect OPD by including limitations to ingestion functions associated with typical development. For this reason, we developed modified cut-points for each of the measures, which we recommend using when assessing preschool children with CP, to improve their specificity.

The PSAS was found to have better specificity than the DDS when calculated using latent-class variable analysis. Despite this, we would support the use of the DDS with modified cut-points as the best available measure for determining OPD in preschool children with CP. The scoreable version of the PSAS is no longer in use, and while the PSAS checklists are useful clinically, there are no cut-points to guide practice decision, and this is less useful for research purposes. The DDS is a measure which is still published; the clinical decisions required for scoring are more specific (which would mean that its reliability would likely remain strong even when used by less familiar users); its scoring structure is more systematic and easily interpreted (that is, each texture covers the same 8 ingestion functions (except for exclusion of the *chewing* item for non-chewable foods and fluids); and an increase in score reflects a linear increase in the extent of the functional impairment.

Substudy 2: Cross-Sectional Study of Oropharyngeal Dysphagia in Children Aged 18 to 36 Months

H^{2A} The prevalence of feeding difficulties will be lower than that reported in the literature (based on a more representative sample of children with CP from all motor severities).

This study found 85% OPD prevalence in preschool children with CP (impairment on 1 or more of the SOMA, DDS, or pharyngeal signs), although this estimate may be lower once accounting for limitations to ingestion functions associated with typical development (45% to 64%). The overall unmodified OPD estimate is consistent with the prevalence estimates reported in the only 2 previous studies conducted in preschool children with CP, of 78%[54] and 90%.[2] The estimate by Reilly et al[2] of 90%, obtained through direct assessments of a community-based sample, used the SOMA alone; a significantly higher estimate compared to the present study estimate using only the SOMA, of 40%. This discrepancy likely reflects the bias towards recruitment of participants with more severe gross motor impairments in the study by Reilly et al (70% had severe to profound motor impairment), when in fact the population distribution of gross motor severity (in developed countries) tends to be skewed to the milder end of the range.[97] The estimate in the study by Wilson et al[54] was based on clinical evidence of oral-motor involvement, which was a broad classification of any neurologically-based impairment of speech subsystems. Tube feeding or cough/ choke/ gag were identified by parent report in close to all of these children (73%). The gross motor severity of children in this study sample was not reported, thus their estimate may also reflect a bias towards recruitment of children with more severe gross motor severities. The current study estimate strengthens previous estimates by using direct clinical OPD measures with strong reproducibility, and a representative study sample across gross motor severity levels.

H^{2B} (a) Children with ambulatory CP (GMFCS I-II) will have only delayed feeding skills, whereas children with nonambulatory CP (GMFCS IV-V) will have delayed and disordered skills.
(b) Clinical signs suggestive of pharyngeal phase impairment will be present across GMFCS levels. Cough will be the most frequent sign in children from GMFCS I-II.

(a) Consistent with this hypothesis, based on the PSAS, about half of the children in GMFCS I-II had delayed oral phase skills (52%), whereas almost all children from GMFCS

IV-V had delayed and disordered skills (100% delayed and 91% disordered). The mean delay score for children from GMFCS I-II was minimal (2 months and 3 months, respectively), which was within the range noted in typical development based on the validation study. The mean delay for children from GMFCS IV-V was much greater, with 13 months and 21 months, respectively; and the disorder scores were 3.7 and 6.6 (out of 9). The surprising finding was that about a fifth of children with ambulatory CP had disordered feeding (23%), which was contrary to the hypothesis. These disorders were minimal and within the range of typical, with a mean disorder score of 0.1 for children from GMFCS I, and 0.4 for children from GMFCS II.

(b) Consistent with the hypothesis, clinical signs suggestive of pharyngeal phase impairment were present in children from each GMFCS level, increasing in number as GMFCS increased. There were 56% and 60% of children from GMFCS I and II with clinical signs, although this was not significantly different from the proportion noted in children with TD (38%). As hypothesised, cough was the most commonly occurring sign in children with ambulatory CP, although this was also frequently noted in children with TD. Once adjusting for the potential that a single cough on thin fluids was part of typical development, 35% of children from GMFCS I were still observed to show clinical signs and 13% of children from GMFCS II.

H^{2C} There will be a negative relationship between OPD prevalence and severity, and gross motor function in children with cerebral palsy aged 18 to 36 months.

There was a negative relationship between OPD prevalence and severity, and gross motor function in our study. There was an increasing number of children with OPD for each increase in GMFCS level, and this difference between groups was statistically significant for each subtype (oral phase, pharyngeal phase and saliva control). Oropharyngeal dysphagia was present across all gross motor severity levels, with as few as 18% of children in GMFCS I identified as having OPD using the SOMA, and as many as 56% of children in this group using the DDS. While the trend for increasing prevalence of OPD with increased gross motor severity was stepwise for each GMFCS level, these relationships were generally only significant for children in GMFCS III-V compared to GMFCS I. Children's mean OPD severity on the DDS increased stepwise as gross motor function declined, from 0.8 (out of a maximum impairment score of 22) in children with TD to 19.1 in GMFCS V (solid scaled score from 0-10 was for TD=0.1, GMFCS V=8.8; fluid

scaled score for TD=0.8, GMFCS V=8.6). Scores were significantly higher for all GMFCS levels when compared to the children with TD (correlation coefficient r=0.89, P < .001).

Substudy 3: Longitudinal Study of Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy

H^{3A} OPD prevalence will be lower at assessment 2 (36 months) compared to assessment 1 (18 to 24 months), owing to a later maturation of oropharyngeal feeding skills in children with CP.

The classification of OPD remained relatively stable between 18 to 24 months and 36 months, when removing classification error based on intra-child variability and limitations to ingestion functions associated with typical development. Consistent with the hypothesis, the prevalence of OPD reduced between assessments. This was only a marginal reduction (3%), however, with all of the improvement noted in children from GMFCS I. Drawing parallels from the gross motor literature,[98] we would expect children with ambulatory CP to have the greatest rate of improvement during the preschool years, whereas children from GMFCS V to plateau earlier. When using the modified cut-points of the DDS, the OPD classification accounts for a degree of typical maturation on the measure scores, so the 8 children who changed classification on the DDS likely reflect the later maturation occurring for children with CP (mostly children from GMFCS I).

H^{3B} GMFCS will be more strongly associated with OPD prevalence at assessment 2 (36 months) compared to assessment 1 (18 to 24 months).

Gross motor function remained the best predictor of OPD classification and severity at both assessment points. That is, there were more children with OPD (on the DDS or pharyngeal signs) as gross motor function declined, regardless of assessment point. Similarly, OPD was more severe with poorer gross motor function regardless of assessment. GMFCS appeared to be more strongly associated with OPD outcomes at the second assessment, although when the relationship was analysed by combined GMFCS level (I-II, III, IV-V), this pattern differed. All children from GMFCS IV-V had OPD (on both the DDS and pharyngeal signs), and their OPD severity was significantly greater than that of children from GMFCS I-II at both assessments. There was not a significantly larger proportion of children from GMFCS III with OPD on the DDS at 18 to 24 months, or OPD

on pharyngeal signs at either assessment. OPD severity was significantly worse for children in GMFCS III at 18 to 24 months (compared to GMFCS I-II), but this was not true for their second assessment at 36 months. Gross motor function was the only risk factor tested which was related consistently to OPD outcomes at both assessment points.

H^{3C} OPD severity at assessment 1 (18 to 24 months) will be strongly associated with poor growth, poor nutritional status, and introduction of nutritional interventions at assessment 2 (36 months).

The only health outcomes which were related to early feeding variables (once accounting for GMFCS and gender) were low weight and BMI, and parent stress. Low weight and BMI were related to the presence of OPD on the DDS at 18 to 24 months (using modified cut-points). This supports the construct validity of the DDS as a measure which is detecting children at risk of later poor nutritional status. Parent stress during mealtimes (at 36 months) was related to the number of challenging behaviours exhibited by their child at 18 to 24 months. OPD on the DDS was not related to this outcome, suggesting it is a child's active resistance to mealtimes which increases the likelihood of stressful mealtimes for parents, rather than the child's motor difficulty during ingestion. Contrary to our hypothesis, OPD severity was not related to any health outcomes.

Substudy 4: Oropharygeal Dysphagia in Preschool Children with Cerebral Palsy Studied in a High-Resource and Low-Resource Country

H^{4A} The prevalence and severity of OPD in Bangladesh will be greater than that reported in the literature.

It was hypothesised that the prevalence and severity of OPD in Bangladesh would be greater than that reported in the literature, owing to a greater prevalence of children from GMFCS III-V, later diagnosis, later and less access to treatment, and cultural factors. Contrary to our hypothesis, the prevalence of OPD in Bangladesh was 68% (based on the DDS with modified cut-points), which is equal or less than reports in the literature (between 19 to 99%). The mean DDS score was 10.4 (SD=7.9), and 16.0 (SD=4.2) for children from GMFCS IV-V. The mean score for children from GMFCS IV-V was comparable to the median score on the DDS for children with CP in a study by Calis et al of 15.0, which also refutes the hypothesis.[1]

H^{4B} The prevalence and severity of OPD and its subtypes will have a strong positive relationship with gross motor function in both countries.

The prevalence of OPD increased with poorer gross motor function for both samples (Australia OR=2.6 (95% CI=1.8, 3.8), *P* < .001; Bangladesh OR=7.3 (95% CI=2.8, 18.8), *P* < .001). This was consistent with our findings in earlier studies, and that of other researchers. GMFCS was the strongest risk factor for OPD across countries, suggesting it is a good predictor of OPD regardless of ethnicity or resourcing.

H^{4C} The prevalence and severity of OPD in children with CP in a low-resource country, when stratified for GMFCS level, will be greater than that in the Australian sample.

The differences in the sample recruitment used in the 2 substudies may have influenced the gross motor severity of the samples, and as such, our a priori analysis plan was to stratify for GMFCS to account for some of these differences. Once stratified for GMFCS, the differences in the presence and severity between countries were minimal (prevalence OR=2.4, *P* = .051; severity β=1.2, *P* = .08). In some instances the proportion of OPD was actually lower in Bangladesh, contrary to our hypothesis. The only statistical differences were fewer children from GMFCS I having OPD on the DDS in Bangladesh, and fewer from GMFCS V having OPD on the pharyngeal phase and on parent report in Bangladesh. This was a surprising finding, when children in Bangladesh have later diagnoses, later and less access to therapy. The OPD severity for children classified as GMFCS IV was significantly lower in Australia (β= -3.5, *P* = .04).

11.2 Clinical Picture of Oropharyngeal Dysphagia according to Gross Motor Function Classification System (GMFCS)

This section combines the results from across each of the substudies, to provide a clinical picture of OPD by GMFCS level. A limitation of OPD research in CP to date is its lack of well defined sample characteristics, which is of particular importance with the heterogeneity of this varied diagnosis. As such, the findings from this research will be discussed with reference to the literature when gross motor function is available or able to be inferred. A summary of OPD subtypes and OPD related factors by GMFCS is shown in Table 5.

Gross Motor Function Classification System I (n=57)

Children who have the potential to independently ambulate have typically received less attention in the OPD literature, perhaps owing to the assumption that the oral sensorimotor mechanism is not impacted in these children. The findings from this doctoral research suggested that about a third of children from GMFCS I had OPD, even once adjusted for limitations to ingestion functions associated with TD.[99] One study reported specifically on OPD prevalence in children with CP (born >35 weeks) from GMFCS I, although this subgroup was small (n=8).[86] This study assessed OPD through direct questioning and observation at 2 years of age. They reported OPD in 25% (n=2) of children, which was comparable to the current research finding. Compared to our sample of children from GMFCS I in Bangladesh, there were significantly greater number of children in Australia with OPD (there were no children from GMFCS I in Bangladesh with OPD on the DDS). This difference may, in part, be explained by the significantly more children in Australia with unilateral spasticity. While not significantly related to OPD outcomes, there was a trend suggesting children with unilateral lesions are more likely to have OPD (of the pharyngeal phase). Considering not only the children's GMFCS level, but also the representation of motor types is important when comparing study findings or generalising to other clinical populations.

The inclusion of children with TD in the present study allowed an understanding of how OPD in children from GMFCS I differed from children of the same age without CP. The mean OPD severity (DDS part 2 raw score) was significantly higher than that of children with TD, although within the modified cut-point range for a determination of OPD. While there were some oral phase impairments which were not significantly different for children from GMFCS I compared to TD (such as *oral transport* of non-chewable and chewable foods, *orienting* for fluid boluses, sipping from cup, and liquid loss), most ingestion functions were impaired in significantly more children with CP. The same was true for many of the clinical signs, except for coughing and wet breathing.

Children from GMFCS I had almost all food/ fluid textures included in their diets, with the exception of tough chewable foods. This modification may reflect family preferences for young children rather than reflecting OPD. This group of children were judged to be safe on all food textures, and only about 10% were recommended for referral for instrumental assessment on thin fluids. Only 3 children from GMFCS I continued to have OPD at 36 months (versus n=8 with OPD at 18 to 24 months), suggesting a later

maturation of oropharyngeal skills in this group. The improvement in OPD severity was only modest (1.6 points, which was within the margin of measurement error). No children from GMFCS I were tube fed and approximately 10% had dietary supplements. Their average energy intake of 4274kJ was adequate for about half of the group (according to population reference values[100]). Preschool children with ambulatory CP have been found to have energy requirements similar to that of their typically developing peers, so the fact that half of the children were not meeting requirments warrants attention.[101] In the study by Sullivan and colleagues, only a small proportion of children with mild CP were reported to have calorie supplements (2.6%, fewer than the current study), gastrostomy feeding (2.6%, more than the current study) and prolonged mealtimes (in 7.9% in their sample).[56] On average, children's nutritional status was within the normal range, with only one child considered underweight (BMI z score< -2).

Gross Motor Function Classification System II (n=15)

Oropharyngeal dysphagia in preschool children functioning at GMFCS II has also received limited exploration in the literature. In a number of our substudies GMFCS I and II were combined to improve the power of the sample, as they are functionally similar levels. It appears that this group, while similar to GMFCS I, also has some distinct differences. The estimates from this group showed poorer precision, in part owing to the smaller sample (n=15), and also related to greater heterogeneity within this subgroup. It was estimated that about 40% of children had OPD (on the modified DDS) and the mean severity for children from GMFCS II was approximately equivalent to the modified cut-point for OPD (3.9). Parents also reported more severe OPD in children from GMFCS II (mean score of 4.1) compared to GMFCS I. One study reported specifically on OPD prevalence in children with CP (born >35 weeks) from GMFCS II, although this subgroup was very small (n=6).[86] This study assessed OPD through direct questioning and observation at 2 years of age finding OPD in 50% (n=3) of children, which was comparable to the current research finding.

A greater range of oral phase impairments observed in over half of the subgroup were noted in GMFCS II compared to GMFCS I (stripping spoon, biting, food loss, cleaning behaviours, chewing, and sipping from a cup). The clinical signs (coughing and wet breathing) impaired in >10% of the subgroup were the same as GMFCS I, although surprisingly impaired in fewer children in GMFCS II. This may, in part, be understood in light of parents' severity scores, which were higher in GMFCS II compared to I. If parents

perceive a greater limitation to children's feeding, they may instinctively be modifying the mealtime, reducing some of the apparent limitations which were observed in GMFCS I. Similar to the GMFCS I subgroup, children from GMFCS II had all foods included in their diets, except 30% who had tough chewable foods excluded. Safety on textures was again the same as children from GMFCS I with all children safe on solids, and about 15% recommended for referral for instrumental assessment of thin fluids. Children's feeding efficiency was marginally lower than GMFCS I, although this did not appear to influence their overall dietary intake relative to average intake of GMFCS I (4434kJ). Sixty percent of children met recommended intake, with a positive mean BMI z score (0.6), and only 1 child considered underweight. There were not any children from GMFCS II using tube feeding, but a greater proportion of this group had modifications to their oral diet (27%). The slightly more pronounced OPD in children from GMFCS II perhaps means that parents and clinicians are more aware of potential difficulties, and are therefore making appropriate modifications to counter negative health outcomes.

A greater number of studies have reported on OPD in children from GMFCS I-II combined as an ambulatory subgroup, or GMFCS I-III combined. One small study of GMFCS I-II (n=10) reported 70% OPD as assessed on VFSS and questionnaire, with piecemeal deglutition (50%), residue in the valleculae (60%) and difficulty with textured food (20%) the most frequently cited concerns.[72] Unlike our sample, coughing was almost nonexistent in these children (10%). Another study of children with CP aged 4 to 16 years found significantly lower scores compared to controls on functional feeding on formal assessment using the FFAm; with spoon feeding, biting and straw drinking within normal limits, and chewing, cup drinking, and swallowing mildly impaired.[38] This trend is consistent with the current study, although direct comparison was not possible with the data available. Sullivan et al combined children with moderate gross motor impairments (with and without mobility aids, judged to be GMFCS II-III), finding high rates of parent-reported choking on food (61%), again, far higher than our estimate of 16%.[56] It is possible that this terminology differed between these studies, with *choking* including coughing, choking and gagging. The number of children with prolonged mealtimes (greater than 3 hours feeding per day) was marginally higher in our research, with 13% reported by Sullivan et al, and 22% in the present study. Conversely, the estimates found in a study of children from GMFCS I-III were lower than the current findings for all reported outcomes except for nasal regurgitation (1%); including difficulty biting (1%), insufficient chewing (1%), difficulty with swallowing (7%), and gagging (6%).[94] Their feeding protocol was not well defined, which may constitute the source of this variability.

Gross Motor Function Classification System III (n=23)

Three out of four preschool children functioning at GMFCS III were found to have OPD (modified cut-points) and their mean severity was 6.8 (maximum of 22). This proportion was within the range reported in the literature for children from GMFCS III, although there was large variability in these estimates from as little as 11%[26] up to 100%.[72] The lower estimate from the study by Waterman and colleagues is limited by poorly defined definitions of OPD and motor severity classifications (moderate motor severity was interpreted as equivalent to GMFCS III). Furthermore, the estimate of 100% by Kim and colleagues was from a very small subgroup of n=7, which reduces confidence in this finding. The range of oral phase impairments which were present in over half of the subgroup was similar to those in GMFCS II, with the addition of impaired oral transport of chewable foods and fluids. This is consistent with a greater proportion of children from GMFCS III also requiring multiple swallows, suggesting more generalised oral phase impairments. One small study of children with CP from GMFCS III found almost 60% of children had poor bolus formation, and 70% with oral residue, piecemeal deglutition, and multiple swallows, supporting this proposition that children at this level are beginning to have more generalised OPD.[72] This was also evidenced by a greater number of children continuing to use infant bottles (22%) and less on open cups (17%).

All children from GMFCS III had purees, semi-solids and thin fluids in their diets, however there were greater numbers who had chewables (14%) and tough chewables (50%) excluded. Similar to GMFCS II, children from GMFCS III all received their nutrition orally, but almost a third had nutritional modifications to their diet. The North American Growth in CP Study (aged 2 to 18 years) found 26% of children from GMFCS III to have modified diets, with 22% requiring chopped or mashed foods, consistent with the finding of the present study, of 14% who did not have chewables in their diet.[55] No children in the study by Fung et al had severe difficulties consuming liquids and foods, which was considered equivalent to the absence of feeding tubes in children from GMFCS III in the present study. The safety of the swallow was questionable for about 10% of children for each texture. Children's energy intake was poorer than that of children from GMFCS I-II on average (3941kJ), and only about a fifth were meeting their energy requirement (based on population data). The influence of this lower energy intake on nutritional status (based on BMI) was not evident, with children from GMFCS III having similar z scores as those in GMFCS I.

Gross Motor Function Classification System IV (n=12)

GMFCS IV was the smallest and also the most heterogeneous GMFCS level. While most children functioning at GMFCS IV had OPD (90%), children from this subgroup had DDS scores ranging from 1-22 (the full range of possible impairment scores). The proportion with OPD was comparable to the estimate of 78% in a previous study of 23 children with CP from GMFCS IV (aged 2 years at assessment), although this study used informal observations of the mealtime.[86] Children from GMFCS IV had a similar range of impairments to ingestion functions to those of children in GMFCS III, but with the addition of difficulty transporting purees as well as chewables and fluids orally, saliva loss during eating, impaired reception of fluids on both cups and bottles, and liquid loss. Children from level IV had the same range of clinical signs observed commonly in children from GMFCS III, but also fremitus (rattly chest) suggesting that food/ fluid or secretions are lower in the respiratory tract. Children with more severe neurological impairments may have a higher rate of silent aspiration and/ or less ability to clear aspirated material despite a similar incidence of cough.[102]

Related to these findings, it is unsurprising that a larger proportion of children (a third for purees and fluids, and a quarter for semi-solids and chewables) were considered to be unsafe (ie, recommended for referral, or exclusion of a texture). All children from GMFCS IV ate pureed foods and drank thin fluids, but semi-solids, chewables and tough chewables were excluded to varying degrees. Half of these children still used an infant bottle as their primary utensil, and only a quarter (n=3) had progressed to open cups. There were 38% of children from GMFCS IV aged 2 to 18 years in the North American Growth Study who experienced difficulty on textures, with 28% requiring mashed/ chopped food (greater than the 10% in our study).[55]

Children from GMFCS IV had the greatest proportion of children improve between assessment 1 and 2 (12 to 18 months later), although there were only 4 children from GMFCS IV in the longitudinal study. Tube feeding was used to supplement oral nutrition in a quarter of children from GMFCS IV (17% using supplementary tube feeds and 8% on predominant tube feeds). Rates of gastrostomy use in our sample were equivalent to those reported in the study by Martinez-Biarge et al for children from GMFCS IV conducted at the same age point.[86] Including the children with tube feeding in the estimate, children's average daily intake was 4084kJ, with only 40% meeting requirements, better than the proportion meeting requirements from GMFCS III. The lowest physical activity level was

used to determine energy adequacy for nonambulatory children aged 3 years, owing to previous findings by our team that their requirements could be up to 30% lower than children with TD.[101] The mean BMI z score of children from GMFCS IV was similar to that of children in GMFCS III (mean= -0.1). This suggests that children from GMFCS III are likely to be more at risk of poor energy intake as they are relying only on oral nutrition, whereas the children from GMFCS IV (being supplemented by tube feeding) are maintaining intake, despite a greater average OPD severity. Parent stress was the highest in GMFCS IV compared to other GMFCS levels (mean of 2.8 out of 5, suggesting mealtimes are moderately stressful).

Gross Motor Function Classification System V (n=23)

All children functioning at GMFCS V had OPD, and this group was more homogenous than others, with mean severity of 19 out of 22 (and confidence range from 18-20). This is consistent with the literature finding that almost all children from GMFCS V (or IV and V) have OPD.[1,62,72,86] A median score on the DDS of 15 was reported in a study of children with CP from GMFCS IV-V, although this sample had fewer children who were not fed orally (15%) which would have made their estimate slightly lower (tube feeding has a default score of 22).[1] Each of the ingestion functions were impaired in greater than 80% of children from GMFCS V, except for *stripping the spoon* (78%), *orienting* to the fluid bolus (79%), and *liquid loss* (61%). About half the children from this level received their fluids via gastrostomy, and furthermore, *liquid loss* would have been reduced due to a higher use of bottles and trainer cups (only 2 children used open cups), and caregivers providing greater control of the volume and pace of fluids.

Most children from GMFCS V had purees in their diets (77%), although these made up a small part of their energy intake overall (on average only 9%). No children were considered safe on any solid texture, and only 18% on thin fluids. The North American Growth Study found that about 80% of children from GMFCS V had feeding dysfunction related to food/ fluid textures, with 40% exhibiting severe difficulties consuming liquids and foods.[55] Consistent with this finding and the present study, nutrition via gastrostomy has been reported in about half of all children from GMFCS V.[86] Children from GMFCS V showed minimal change after 18 to 24 months. This subgroup appeared to have reached their ceiling of performance for purees by 18 to 24 months (with all children impaired on all items, and no change between assessments). More children from GMFCS V showed impairment on ingestion functions for chewable foods at 36 months compared to 18 to 24

months, perhaps due to the introduction of firmer chewable foods between these ages for children from GMFCS V. Children's average daily energy intake was much lower than that of other GMFCS levels (3237kJ, with only 12% meeting requirements), although this may reflect differences in energy requirements (owing to their smaller size and lower activity) rather than inadequacy.[101] Children in GMFCS V were much smaller than the other GMFCS levels (BMI z score= -0.7), with 5 out of 23 (22%) being underweight.

Limitations and Implications of the Doctoral Study

The prevalence estimate and findings from this study are thought to be a plausible reflection of that of the general population of CP. A representative population-based sample was recruited in this study, and these children were assessed using direct standardised clinical assessments. Furthermore, the use of the GMFCS as a universally used motor severity classification, against which all of the OPD outcomes were described, allows these findings to be generalised to any given population, regardless of the gross motor function distribution. This was demonstrated by our findings in Bangladesh, which showed equivalent OPD prevalence and severity once stratified for GMFCS.

Limitations

Despite these study strengths, there were a number of potential limitations which influenced the analyses that could be performed, or the findings of the research:

i. The greatest limitation to the field of OPD research is the lack of a gold standard measure of OPD, and a lack of consensus regarding the domains encompassed by the term. Our measure selection was informed by the clinimetric review (Chapter 2), however, each measure had limitations associated with the construct it captured, its psychometric properties, score stability (particularly between children's mealtimes), interpretation of scores, or lack of validation against a typically developing sample or health outcomes. The field of paediatric dysphagia may also continue to lack a single 'gold standard measure', owing to the broad range of feeding problems associated with neurological diagnoses.

Table 7. Clinical Picture of Oropharygneal Dysphagia and Related Factors, Based on Gross Motor Function (GMFCS)

	GMFCS I (n=57)[a]	**GMFCS II (n=15)[a]**	**GMFCS III (n=23)[a]**	**GMFCS IV (n=12)[a]**	**GMFCS V (n=23)[a]**
Prevalence on DDS					
Standard (%)	66.7	100.0	95.7	100.0	100.0
Modified (%)	30.9	42.9	71.4	90.0	100.0
OPD severity (mean, CI)	2.4 (1.6, 3.1)	3.9 (1.9, 5.8)	6.8 (4.9, 8.6)	12.1 (7.7, 16.5)	19.1 (18.0, 20.3)
Oral phase impairments[b]					
Solids	Cleaning	Stripping spoon Biting Food loss Cleaning Chewing	Stripping spoon Biting Food loss Cleaning Oral transport (solid) Chewing	Stripping spoon Biting Saliva loss in eating Food loss Cleaning Oral transport (puree) Oral transport (solid) Chewing	Orienting Stripping spoon Biting Saliva loss in eating Food loss Cleaning Oral transport (puree) Oral transport (solid) Chewing
Fluids	nil	Sipping from cup	Sipping from cup Oral transport	Stripping bottle teat Sipping from cup Liquid loss Oral transport	Orienting Stripping bottle teat Sipping from cup Liquid loss Oral transport
Clinical signs[c]	Coughing Wet breathing	Coughing Wet breathing	Coughing Multiple swallows Gurgly voice Wet breathing	Coughing Multiple swallows Gurgly voice Wet breathing Rattly chest	Coughing Multiple swallows Gurgly voice Wet breathing Gagging Rattly chest

	GMFCS I (n=57)[a]	GMFCS II (n=15)[a]	GMFCS III (n=23)[a]	GMFCS IV (n=12)[a]	GMFCS V (n=23)[a]
(clinical signs, continued)					Respiratory effort
					Respiratory rate
					Snuffly nose
					Eye tearing
					Colour change
Saliva control (%)	38.6	26.7	47.8	75.0	87.0
Challenging behaviours (mean, CI)	6.5 (5.3, 7.7)	6.8 (4.5, 9.2)	5.5 (3.6, 7.4)	8.3 (5.7, 11.0)	5.6 (3.5, 7.7)
Parent-report prevalence (%)	71.9	66.7	78.3	91.7	100.0
Parent-report severity (0-20)	2.0 (1.3, 2.7)	4.1 (0.7, 7.4)	5.6 (2.8, 8.5)	7.7 (3.6, 11.7)	15.8 (13.5, 18.0)
Textures included (%)					
Puree	95.6	100.0	100.0	100.0	76.5
Semi-solid	93.3	92.3	100.0	90.0	35.3
Chewable	100.0	100.0	85.7	90.0	35.3
Tough chewable	88.9	69.2	50.0	60.0	5.9
Fluid	100.0	100.0	100.0	100.0	41.2
Thickened fluid	0.0	0.0	0.0	0.0	5.9
Primary fluid utensil					
Bottle	5.3	0.0	21.7	50.0	13.6
Trainer cup	50.9	53.3	56.5	25.0	22.7
Cup	43.9	46.7	17.4	25.0	9.1
Safe swallow[d] (%)					
Puree	100.0	100.0	92.3	62.5	0.0
Semi-solid	100.0	100.0	91.7	71.4	0.0
Chewable	100.0	100.0	91.7	71.4	0.0
Thin fluid	90.7	83.3	90.9	66.7	18.2
Efficiency (mean g/minute)	9.0±5.4	7.9±3.5	9.9±10.9	13.5±17.5[g]	7.7±3.7[g]
Duration (mean minutes/ day)[e]	141.2±10.1	135.4±13.4	140.6±91.4	120.3±65.3	155.5±19.8

	GMFCS I (n=57)[a]	GMFCS II (n=15)[a]	GMFCS III (n=23)[a]	GMFCS IV (n=12)[a]	GMFCS V (n=23)[a]
Maturation (mean DDS score change)	1.6 (-0.2, 3.4)	2.2 (1.2, 3.1)	2.3 (0.7, 3.9)	4.3 (0.9, 7.6)	1.3 (-0.9, 3.5)
Gastro-Oesophageal Reflux	5.8	15.4	8.7	27.3	47.6
Supplementary nutrition (%)					
Total oral	89.5	73.3	65.2	41.7	13.0
Total oral with modifications	8.8	26.7	30.4	33.3	30.4
Supplementary tube-feeds	0.0	0.0	0.0	8.3	8.7
Predominately tube-feeds	0.0	0.0	0.0	16.7	21.7
Total tube-feeds	0.0	0.0	0.0	0.0	21.7
Energy intake (mean kJ)	4274.2±119.2	4434.7±208.1	3941.7±430.7	4084.6±434.3	3237.2±212.8
Adequate energy intake[f] (%)	51.1	61.5	21.4	40.0	11.8
BMI z score (mean, CI)	0.1 (-0.2, 0.4)	0.6 (-0.1, 1.3)	-0.2 (-0.8, 0.4)	-0.1 (-1.1, 1.0)	-0.7 (-1.4, 0.1)
Hospitalisation for LRTI (%)	13.6	0.0	9.1	0.0	50.0
Parent stress, 1-5 (mean, CI)	1.8 (1.6, 2.1)	1.7 (1.4, 2.1)	1.8 (1.3, 2.3)	2.8 (2.3, 3.2)	1.9 (0.7, 3.0)

Abbreviations: BMI, Body Mass Index; CI, Confidence Interval; DDS, Dysphagia Disorders Survey; g, grams; GMFCS, Gross Motor Function Classification System; kJ, kilojoules; LRTI, Lower Respiratory Tract Infection

[a] Participant numbers reflect total GNPA sample, but numbers varied between studies.
[b] Present in >50% of level.
[c] Present in >10% of level.
[d] Safe includes *continue* and *supervision* recommendations, unsafe includes *referral* and *exclude* recommendations.
[e] GMFCS IV-V exclude tube fed children.
[f] Requirement based on Australian National Health and Medical Research Council Nutrient Reference Values.[100]
[g] Includes only completely orally fed children.

ii. There were 130 children recruited to the study (for their initial appointment between 18 and 36 months), and an additional 40 children with TD (serving as the reference group), which was adequate to provide precision to our overall prevalence estimates and allow comparison between GMFCS levels. Larger numbers, however, would have allowed better exploration of OPD according to each GMFCS level, particularly for the midrange (GMFCS II-IV), which are both smaller in number, and more heterogenous. To account for this, in some of the substudies, GMFCS levels were combined into ambulatory (GMFCS I-II), ambulatory with assistance (GMFCS III) and nonambulatory (GMFCS IV-V). In addition, the small numbers of children without spasticity (ie, dyskinesia, hypotonia, and ataxia), which is reflective of the population distribution of motor type, reduced our ability to explore relationship between motor type and OPD, apart from the combined Bangladesh-Australia sample. Limitations to these subtype analyses may only be resolved through multi-site or register studies.

iii. The sample of children with TD was recruited specifically to validate the standardised measures of OPD. As such, we did not have prospective TD data for all of the study variables, such as dietary intake, feeding efficiency, challenging behaviours, and health outcomes. While there are normative data available for some of these variables in the research literature (such as energy intake), TD data would have contributed to our understanding of the OPD factors which are significantly different for children with ambulatory CP.

iv. The study was conducted over the course of a 4 year period (beginning in April 2009), and as such, live ratings of the mealtime by a speech pathologist were not possible. Mealtime rating from video is considered an appropriate method for the standardised measures (DDS, SOMA and PSAS), as outlined in the SOMA manual.[103] In the case of the clinical signs suggestive of pharyngeal phase impairment, it is expected that some of these signs may have lacked accuracy when rated from video (particularly respiratory related variables). This factor was unable to be considered when assessing potential sources of difference between direct assessment and parent report for the clinical signs.

v. To maintain the naturalistic component of the mealtime and increase compliance in this young age group, parents were asked to bring their foods (according to standardised texture categories) and utensils from home for the mealtime assessment. This meant that some of the variability seen in OPD patterns between children and mealtimes (including in the test-retest reproducibility and OPD in Bangladesh sub-

studies) may be related to these external factors rather than intrinsic child factors (oral sensorimotor system, child's alertness, motivation).

vi. Other internal and external child factors, such as cognition and appetite, which may influence OPD, were not measured as they were beyond the scope of the study. We cannot, therefore, account for the full range of influencing factors on children's OPD or the relative contribution of those variables associated with gross motor function.

vii. Total scores were not available for all children who completed a mealtime video due to some missing food/ fluid textures. The mealtime assessment may have missed textures for a number of reasons, including refusal by the child, safety on that texture, or parents forgetting to bring the texture to their assessment. Default scoring was available for textures excluded for safety reasons. The omission due to parents not bringing the texture was resolved midway through the 18 to 36 month data collection period by having foods available on stand-by. Data collection forms were also modified to prompt the research assistants collecting the mealtime video to record the reason for omissions. Because some of this data were collected prior to the beginning of the doctoral candidature, we were unable to reliably report on these patterns of omission, which could reflect important data.

viii. Data on the specific treatments children received over the duration of the study were not collected as part of the study protocol (only general information was collected, regarding access to speech pathology). This may have influenced the patterns of OPD; particularly the rate of change in the longitudinal substudy, and differences in access to therapy between countries in the high- and low-resource comparison.

ix. It was not feasible in this study to have all children who were identified at risk of aspiration referred for instrumental assessment (VFSS). We instead reviewed data from VFSS performed during the study period as part of standard clinical management, for which only 9 children had evaluation results available. These numbers were insufficient for statistical analysis which meant we were unable to use this gold standard method for aspiration to validate the clinical signs suggestive of pharyngeal phase impairment.

Clinical Implications of the Doctoral Study

The findings of this research provide important information to guide clinical practice:

i. Central to this doctoral research was the exploration of the clinimetric properties and usefulness of measures of OPD. As such, this study has provided clinicians with

detailed information regarding the best measure to use for different purposes, and guidance on interpretation of the results. Part of this exploration was the addition of typically developing reference data for each of the standardised measures and clinical signs.

a. The psychometric properties of a measure are often less valued in the clinical context than the usefulness of the data to guide decision making. While we advocate the use of the DDS as the best measure to use for research, it may be that the PSAS is a more preferred choice for use in a clinical context due to it providing comprehensive age-referenced data. The DDS and PSAS performed similarly in their reproducibility and validation, and it appears both are measuring a similar construct. For its use in this study, the original PSAS form was updated to make it more user friendly (while not altering the wording or structure, Appendix 21).
b. All 16 signs used in this research had strong reproducibility for interrater and intrarater. The validity of the signs in discriminating possible pharyngeal dysphagia from signs associated with typical development is important when interpreting their use. This research found a single cough on thin fluids was present in many children with TD, as well as being variable between mealtimes. As such, the isolated use of one cough as a marker of OPD in children with CP, aged less than 36 months, should be employed with caution. Furthermore, assessing clinical signs in only a single mealtime may not accurately describe the child's OPD, so training parents to detect clinical signs across a number of mealtimes may be a more feasible and accurate method.

ii. Earlier identification of children with OPD through improved screening:

a. The findings provide guidance on the use of parent report as a feasible method for screening. Parent-reported ability on textures was not a strong measure of OPD, it was reasonable for detection of children which were identified as having safety concerns using direct assessment. Providing parents with more specific screening questions yielded stronger agreement with direct assessment. There is a need to develop resources, such as training video clips, to support parents' detection of specific oropharyngeal impairments, in particular observation of clinical signs suggestive of pharyngeal phase impairment.
b. GMFCS was a powerful predictor of OPD, and this relationship remained for OPD subtypes, varying children's ages, ethnicity and different resourcing. As such, the GMFCS may be a useful system to guide OPD screening in preschool children with CP. This classification is widely used by various clinical disciplines, and has been

shown to be stable across time (from early years to adolescence). The EDACS (Eating and Drinking Ability Classification System) may also provide a useful adjunct classification to the GMFCS, although its validity and long term prognostic characteristics are yet to be tested, particularly in young children.

iii. This research has provided important data to assist clinicians to counsel parents regarding their child's feeding and nutritional prognosis. While the longitudinal data presented in this study were only across an 18 month period, patterns of maturation and change can begin to be elucidated. Furthermore, the detailed descriptions of OPD in each GMFCS level used in conjunction with the stability of the GMFCS across time, will assist in educating parents about their child's expected presentation and progression of skills. This data will also help clinicians and researchers to plan and prioritise more appropriate therapeutic and nutritional interventions.

iv. Mealtime safety, efficiency and dietary adequacy should all be considered when making mealtime decisions. This study found children with poorer gross motor function were increasingly reliant on fluids for meeting their nutritional needs. Thin fluids have frequently been documented as the texture which carries the greatest safety risk, including a greater proportion of clinical signs observed on this texture in the current research. Thin fluids may also be the texture most efficiently consumed by children, particularly those with poorer gross motor function. This presents an important tension for feeding clinicians to consider when making supplementary nutritional and mealtime recommendations, between safety and efficiency of dietary intake.

v. A child's feeding efficiency across mealtimes (distinct from the feeding efficiency described by Gisel on a single bolus) may be a useful tool for decision-making in conjunction with other OPD measures. This measurement accounts not only for the reduced efficiency associated with oromotor compromise, but also that associated with sensory related compromise, such as challenging behaviours and mealtime avoidance.

vi. The measure of habitual texture consumption (ie, percentage of their diet constituted by each food/ fluid texture) may provide useful information to assist clinicians in understanding not only the presence of a texture which may be unsafe or inefficiently consumed, but also how frequently the child is being exposed to that risk. Further validation would be useful to determine the minimum length of time necessary to record to gain an accurate assessment. This may be a useful measure for both clinical use and research.

Research Implications and Recommendations for Future Research

The results of this doctoral thesis will contribute towards enhanced targeting of research in the field of OPD in children with CP. Some specific areas to consider when planning research in paediatric feeding would include:

i. The choice of standardised assessment should be considered when embarking on research in OPD, nutrition or related fields. All 3 measures used in this thesis (SOMA, DDS, and PSAS) had strong agreement between raters for detecting OPD, and so one measure should not be prioritised for use based on this property. The PSAS and DDS were in agreement regarding case status about 85% of the time, which suggests they are both capturing a similar construct of OPD. To ensure we are detecting the extent of the impairment, the DDS is recommended as a measure with greater sensitivity. Using our modified cut-points for children aged 18 to 36 months will improve the measure's specificity. Furthermore, the DDS is considered useful as it continues to be published, the clinical decisions required for scoring are more specific (which would mean that its reliability would likely remain strong even when used by less familiar users), and its scoring structure is more systematic and easily interpreted.
ii. Parent report has been used extensively in previous research on OPD, although there has been limited understanding of the accuracy of this as a proxy for a direct objective assessment. This doctoral research found that parents were generally in agreement with direct assessment about half of the time, although this improved when considering agreement with safety on a texture. Parents were not found to consistently overdetect or underdetect OPD presence or severity, as had been previously suggested in the literature. Its use as a measure of OPD in research should be interpreted cautiously, as it may encompass a broad construct, particularly if questions are not well defined.
iii. The GMFCS proved a powerful predictor of OPD prevalence and severity in this study, and was significant across time and contexts. These findings support the continued use of the GMFCS as a framework against which to describe OPD (and other functional variables). During the course of this doctoral research, an increasing number of studies have reported on OPD outcomes against the GMFCS, and this has allowed greater comparison of findings between studies and synthesis of results.
iv. The detailed description of OPD prevalence, severity, and patterns according to GMFCS and longitudinally has contributed a strong foundation for planning future intervention studies. For example, it may be that children with ambulatory CP (GMFCS

I-II) and OPD are more suited to oral sensorimotor treatments, those who are assisted ambulators (GMFCS III) to modifications to textures, utensils and environmental modifications, and those with nonambulatory CP (GMFCS IV-V) better targeted for nutritional interventions. It should also be noted that this distinction is somewhat arbitrary; there were children from GMFCS IV who were functioning more like a child from GMFCS III as far as their oropharyngeal sensorimotor skills. The results from this doctoral research should be considered, in conjunction with emerging findings using the EDACS, when planning intervention studies.

With consideration of these research implications, a number of directions for future research are proposed:

i. A high priority for the progression of both OPD research and clinical management would be the development of a new measure that is sensitive enough to detect changes from interventions/ maturation, and stable enough to reduce the impact of variability in children's skills between mealtimes. Ideally this measure would not be a series of binary judgments of the integrity of the ingestion function summed to give an overall severity (as is the structure of the DDS), but comprise an ordinal scale within each item to detect the severity of impairment for each ingestion function. Areas of the DDS requiring greater detail are the pharyngeal phase, mastication patterns, as well as inclusion of scoring for different fluid utensils. A factor analysis of items from all 3 measures may prove a useful methodology in the development of a new measure.
ii. Whether developing a new measure, or improving the existing measures, further validation is required, particularly to explore the sensitivity of measures. Use of an expert speech pathology panel may serve as the most appropriate gold standard, as well as further testing around health outcomes. A consensus statement regarding the construct of OPD would unite future research efforts, allowing better synthesis of results.
iii. Studies have explored the sensitivity and specificity of a number of clinical signs of aspiration, however, further testing of this in children with CP would be useful. Exploration of other objective measures of aspiration which are accessible outside tertiary care (such as cervical auscultation) could enhance the accuracy of diagnosis. Improving the accuracy of detecting aspiration is only one consideration. Longitudinal studies are needed to determine the effect of chronic aspiration on respiratory health in children with CP and the factors affecting this. Habitual texture consumption may be a useful measure to understand if there is a relationship between the frequency of

exposure to a texture risk and the development of aspiration pneumonias. Alternative diagnostic outcome measures need to be developed and tested to indicate compromise to respiratory health rather than only aspiration on VFSS.

iv. The Eating and Drinking Ability Classification System (EDACS) is a promising classification which may assist in standardisation of research in the field. The application of direct clinical measures (such as the DDS) against the EDACS would provide some interesting insights into both of these measures. In addition, mapping the EDACS against the GMFCS would strengthen this new classification, particularly in light of the findings from the present study.

v. Feeding efficiency across mealtimes was used in the current study to explore a potential factors influencing adequacy of dietary intake. This may be a useful measurement for decision making in conjunction with other OPD measures in future research around the nutritional aspects of OPD. Validating this measure against a direct assessment would be useful.

vi. It was noted in this study that parents were not systematically biased towards overreporting or underreporting, although direct assessment agreed with parent report only about half the time. Parent report presents a useful method for detecting OPD in both clinical and research contexts. As such, further exploration about what the factors parents consider to constitute OPD would assist in its use as a proxy for direct assessment.

vii. The standardised assessments were all focused primarily on the motor aspects of OPD. Little is known about the role of sensory aspects for OPD in children with CP. Studies exploring the sensory aspects of OPD in greater detail would help us understand the mealtime challenges (and oropharyngeal sensorimotoror impairments) associated with a diagnosis of CP.

viii. The longitudinal data gathered from this doctoral research were novel and useful in understanding OPD prognosis in young children with CP. The follow-up of children for a longer time period would contribute to understanding the later health impacts of early OPD.

Conclusions

This thesis explored the prevalence and patterns of OPD in preschool children with CP, according to GMFCS. Its relationship to dietary intake and nutritional status was also examined. The key findings from the thesis were:

i. The validity and reproducibility substudy had 2 key outcomes:
   a. A systematic review of the clinimetric properties of OPD measures found that the SOMA and FFAm were the measures with the best psychometrics, and SOMA and DDS had the best clinical utility.
   b. Testing of the psychometric properties, including validation against a typically developing reference sample suggested the DDS was the most useful measure in research. The modified cut-points should be used when assessing children aged 18 to 36 months.

ii. The cross-sectional studies found that OPD is prevalent in about 60% of preschool children with CP, and is present even in children with ambulatory CP. OPD prevalence and severity were significantly related to gross motor function.

iii. The longitudinal substudy showed that there was minimal change in OPD classification or severity between 18 to 24 months and 36 months. OPD on the DDS (modified cut-points) at 18 to 24 months was related to lower weight and BMI z scores at 36 months. GMFCS was the only risk factor related to all OPD outcomes and across both assessment points.

iv. The comparative study exploring OPD prevalence and severity in a high-resource and low-resource country showed the generalisability of our research findings. Motor distribution (GMFCS) differed significantly between countries which meant that OPD was more prevalent in the Bangladesh sample overall. Once stratified for GMFCS, however, OPD prevalence and severity did not differ significantly between countries.

This thesis supports the proposition that OPD is present in the majority of children with CP, and across all levels of gross motor function. A greater awareness of OPD is needed, particularly in children with ambulatory CP, as it may be frequently overlooked by both parents and clinicians. This thesis has provided useful information as a basis for earlier identification of children with CP who are at risk of growth or respiratory consequences associated with OPD, as well as in planning optimal oropharyngeal sensorimotor therapies and nutritional interventions.

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Appendices

Appendix 1-6: Ethics Approvals and Amendments for Growth Nutrition and Physical Activity study

Appendix 1. Royal Children's Hospital and Health Service District Ethics Committee Approval: HREC/08/QRCH/112
Appendix 2. The University of Queensland Institutional Approval Form for Experiments on Humans Including Behavioural Research Approval: 2008002260
Appendix 3. Ethics amendment: Access to videofluoroscopy results (August 2011)
Appendix 4. Ethics amendment: Additional food textures; Recruitment of TD sample (n=40); Recruitment of reproducibility sample, aged 18-24 months (n=20) (March 2012)
Appendix 5. Ethics amendment: Reporting safety information to parents (November 2012)
Appendix 6. Ethics amendment: Recruitment of reproducibility sample, aged 30-36 months (n=6) (February 2013)

Appendix 7-11: Ethics Approvals and Amendments for Bangladesh Sub-study

Appendix 7. Children's Health Services Queensland Human Research Ethics Committee Approval: HREC/13/ARCH/69
Appendix 8. The University of Queensland Institutional Human Research Ethics Approval: 2013000625
Appendix 9. Centre for the Rehabilitation of the Paralysed Ethics Approval CRP/RE/0401/55
Appendix 10. International Centre for Diarrhoeal Disease Research, Bangladesh (icddr,b) Research Review Committee Approval PR-13047
Appendix 11. Ethics amendment: Electronic versions of study documents stored in Australia (December 2013)

Appendix 12-14: Parent/ guardian information statements and consent forms

Appendix 12. Queensland CP Child: Growth Nutrition and Physical Activity
Appendix 13. Oropharyngeal Dysphagia Reproducibility Substudy
Appendix 14. Oropharyngeal Dysphagia Validity Substudy (Children with Typical Development
Appendix 15. Oropharyngeal Dysphagia in Bangladesh Substudy

Appendix 16-19: Recruitment Flyers

Appendix 16. Queensland CP Child: Growth Nutrition and Physical Activity

Appendix 17. Oropharyngeal Dysphagia Reproducibility Substudy

Appendix 18. Oropharyngeal Dysphagia Validity Substudy (Children with Typical Development)

Appendix 19. Oropharyngeal Dysphagia in Bangladesh Substudy

Appendix 20-21: Evaluation forms and questionnaires

Appendix 20. Queensland Cerebral Palsy Child: Growth Nutrition and Physical Activity Snack protocol

Appendix 21. Oropharyngeal Dysphagia Battery (data sheet, Schedule for Oral Motor Assessment, Dysphagia Disorders Survey, Pre Speech Assessment Scale, checklist of signs suggestive of pharyngeal phase impairments)

Appendix 22. Conference presentations, invited speaker and awards during candidature

Appendix 23-24. Supporting protocols

Appendix 23: Queensland Cerebral Palsy Child: Growth, Nutrition and Physical Activity Protocol (NHMRC 569605)

Bell KL, Boyd RN, Tweedy SM, Weir KA, Stevenson RD, Davies PSW. A prospective, longitudinal study of growth, nutrition and sedentary behaviour in young children with cerebral palsy. *BMC Public Health.* August 27, 2012 2010;10:e179-e191.

Appendix 24: Queensland Cerebral Palsy Child: Brain Structure and Motor Function Protocol (NHMRC 465128)

Boyd RN, Jordan R, Pareezer L, et al. Australian Cerebral Palsy Child Study: protocol of a prospective population based study of motor and brain development of preschool aged children with cerebral palsy. *BMC Neurol.* February 14, 2014 2013;13(57):e57-e69.

ROYAL CHILDREN'S HOSPITAL & HEALTH SERVICE DISTRICT ETHICS COMMITTEE

Professor John Pearn (Chair) 3365 5323
Mrs Amanda Smith (Ethics Officer) 3636 9167

Queensland Government

Queensland **Health**

Level 3, RCH Foundation Building
Royal Children's Hospital
Herston QLD 4029 Australia
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3rd December 2008

Dr Kristie Bell
Dietician/Clinical Postdoctoral Research Fellow
Queensland Cerebral Palsy & Rehabilitation Centre
Royal Children's Hospital & Health Service District
Herston QLD 4029

Dear Dr Bell,

The Queensland Cerebral Palsy Child Study – Growth, Nutrition and Physical Acitivty.

Many thanks for your letter of the 17th November together with the application for Ethics approval for the above project.

This was tabled and reviewed at our meeting on the 1st December and the Committee are happy to give their approval for this work.

An ethics number will be sent to you as soon as possible.

Please do not hesitate to contact me should you have any queries.

With kindest regards,

Professor John Pearn
Chair
Royal Children's Hospital and Health Service District Ethics Committee

Cc: Ethics Committee files (Professor John Pearn)
Members of the Ethics Committee

At a meeting of the Royal Children's Hospital and Health Service District Ethics Committee held on [1st December 2008], the Committee reviewed the above Protocol. It is advised that Royal Children's Hospital and Health Service District Ethics Committee have approved your request for ethical approval.

During the conduct of the study you are required to adhere to the following conditions:

The Royal Children's Hospital & Health Service District Ethics Committee complies with all National Health and Medical Research Council (NHMRC) series of guidelines in accordance with the *National Statement on Ethical Conduct in Human Research 2007,* the *Guidelines under Section 95 of the Privacy Act 1998* and the *Guidelines approved under Section 95A of the Privacy Act 1998.*

1. This letter gives Ethics approval for your project. Approval for research is a two-step process. The second step requires Institutional approval from the District Executive Committee of the Royal Children's Hospital and Health Service District who considers each application, after Ethics approval is given. **Research can not commence until this is obtained.**

2. We require an annual progress report (or sooner if the project is completed) concerning the study. This must include progress to date or outcome in the case of completed research. (In accordance with National Statement 5.5.3)

3. In accordance with the National Statement (3.3.12), before beginning the clinical phase of the research, researchers should register clinical trails in a publicly accessible domain.

4. If the project does not proceed, the Committee must be informed as soon as possible. (In accordance with National Statement 5.5.6)

5. The Committee must be informed of any potential or realised problem with bioethical implications, if such occurs during the conduct of the research project.

5. Any serious adverse event (SAE) that arises in the context of this research, or involving a researcher conducting this research, must be reported to the Ethics Committee within 72 hours and reported to the sponsor (if applicable) within the stipulated time frame.

   Serious Adverse Event Reports that are generated off-site during multi-centre trials are required to be submitted to the Chair of the RCH & HSD Ethics Committee on receipt by the researcher. A summary of the SAE reports is to accompany the submission. Information required includes; patient details (age & sex), adverse event, outcome and the likelihood of the event being related to the study drug/device/procedure.

   With respect to all SAEs, the researcher must provide his or her opinion as to whether the SAE is directly related to the research intervention.

   A copy of the SAE Summary must be provided. (This can be obtained from the Ethics Officer)

6. The Ethics Committee will conduct a randomly identified audit of a proportion of research projects approved by the Committee. That audit process will look at such issues as;
   a. Security of Documents
   b. Consent Form Register
   c. Serious Adverse Events Register
   d. Withdrawal of Participants – who and why
   e. The de-identification of data

7. We require researchers to give a declaration of intention to publish their findings in a refereed journal or similar peer-reviewed forum.

   Your work must be in accordance with the following:

   - National Statement on Ethical Conduct in Human Research: http://www.nhmrc.gov.au/publications/synopses/_files/e72.pdf
   - Queensland Health Management Research Policy: http://www.health.qld.gov.au/cpic/documents/ethics/research_policy.pdf
   - Joint NHMRC / AVCC Statement and Guidelines on Research Practice (1997): http://www.nhmrc.gov.au/funding/policy/researchprac.htm
   - Declaration of Helsinki: http://www.health.qld.gov.au/ethics/Documents/24938_policy.pdf
   - Guidelines under Section 95 of the Privacy Act1995 and Guidelines approved under Section 95A of the Privacy Act 1995. http://www.comlaw.gov.au/ComLaw/Legislation/ActCompilation1.nsf/0/B471AB909A18D172CA25725C0083858A/$file/Privacy1988_WD02HYP.pdf
   - Queensland Health Privacy Guidelines IS42 & IS42A: http://qheps.health.qld.gov.au/privacy/resources.htm

8. The Committee wishes you well with your research. Please contact our office if you require any assistance.

THE UNIVERSITY OF QUEENSLAND

Institutional Approval Form For Experiments On Humans Including Behavioural Research

Chief Investigator: A/Prof Peter Davies

Project Title: Queensland Cerebral Palsy Child - Growth, Nutrition And Physical Activity

Supervisor: None

Co-Investigator(s): A/Prof Roslyn Boyd, Dr Kristie Bell, Professor Richard Stevenson, Dr Sean Tweedy, Ms Kelly Weir, Dr Robert Ware, Ms Harrison Carly, Dr Lynne McKinlay, Dr Kate Sinclair, Mr Michael Delacey, Ms Leanne Jack, Mrs Megan Kentish

Department(s): Children's Nutrition Research Centre, UQ

Project Number: 2008002260

Granting Agency/Degree: NHMRC

Duration: 31st December 2014

Comments:

Expedited review on the basis of approval from the Royal Children's Hospital HREC, dated 03/12/2008.

Name of responsible Committee:-
Medical Research Ethics Committee
This project complies with the provisions contained in the *National Statement on Ethical Conduct in Human Research* and complies with the regulations governing experimentation on humans.

Name of Ethics Committee representative:-
Professor Bill Vicenzino
Chairperson
Medical Research Ethics Committee

Date: 17-12-07 Signature: _____

QLD CHILDREN'S HEALTH SERVICES (RCH)
HUMAN RESEARCH ETHICS COMMITTEE

Professor John Pearn (Chair) 3365 5323
Mrs Amanda Smith (Co-ordinator) 3636 9167

Queensland Government

Queensland **Health**

Level 3, RCH Foundation Building
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Herston QLD 4029 Australia
Telephone (07) 3636 9167
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10th August 2011

Ms Katherine Benfer
PhD Scholar/ Speech Pathologist
Queensland Cerebral Palsy & Rehabilitation Centre
Royal Brisbane Women's Hospital
Level 7, Block 6
Herston QLD 4029

Dear Ms Benfer,

HREC Reference number: HREC/08/QRCH/112
Project title: The Queensland Cerebral Palsy Child Study - Growth, Nutrition and Physical Activity.

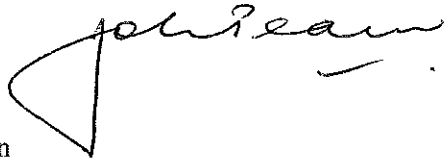
Many thanks for your email with the attached letter dated 27th April 2011, and received here on the 22nd November 2011.

One notes that the research team wishes to extend the above study by reviewing medical files of children with Cerebral Palsy, referred for potential consideration for videofluroscopic swallow studies.

One is happy to approve this extension on behalf of the Ethics committee with one extra point to be considered. The permission of the clinician, in whose name the child is being managed for decisions about such radiographic studies, should be obtained prior to perusal of such charts. In this case, I anticipate that such clinicians would most likely to totally agree with such an approach. If you contacted the Gastroenterologists making such decisions about the videofluroscopic studies, I would anticipate that they would give block approval for access to their clinical records and charts.

With best wishes for the study.

Yours sincerely,



Professor John Pearn
Chair
Queensland Children's Health Services (RCH) Human Research Ethics Committee

Cc: Ethics Committee Files

QLD CHILDREN'S HEALTH SERVICES (RCH)
HUMAN RESEARCH ETHICS COMMITTEE

Professor John Pearn (Chair) 3365 5323
Mrs Amanda Smith (Co-ordinator) 3636 9167

Queensland Government

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Herston QLD 4029 Australia
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Facsimile (07) 3365 5455

1st March 2012

Ms Katherine Benfer
NHMRC PhD Scholar/Speech Pathologist
QIMR
Level 4, Foundation Building
Herston QLD 4029

Dear Ms Benfer,

HREC Reference number: HREC/08/QRCH/112
Project title: The Queensland Cerebral Palsy Child Study - Growth, Nutrition And Physical Activity.
Amendment number: HREC/08/QRCH/112/AM04

Many thanks for your letter dated 17th February regarding an amendment to the above study. This has now been reviewed. I am pleased to advise that the amended documents reviewed and approved were:

Document	Version	Date
Covering Letter		17 February 2012
Protocol		
Letter of invitation to participant – Children with Cerebral Palsy (18-24 months)		
Parent/Guardian Information Sheet and Consent Form (Form E)	1	22 February 2012
Letter of invitation to participant – Typical Developing Children (18-36 months)		
Parent/Guardian Information Sheet and Consent Form (Form D)	1	22 February 2012
Parent/Guardian Information Sheet and Consent Form (Form B)	4	22 February 2012
Parent/Guardian Information Sheet and Consent Form (Form A)	4	22 February 2012

The QLD Children's Health Services (RCH) Human Research Ethics Committee (HREC) is constituted and operates in accordance with the National Health and Medical Research Council's *"National Statement on Ethical Conduct in Human Research (2007), NHMRC and Universities Australia Australian Code for the Responsible Conduct of Research (2007)* and the "*CPMP/ICH Note for Guidance on Good Clinical Practice"*.

A copy of this letter must be forwarded to the Research Governance Officer. It should be noted that all requirements of the original approval still apply.

Yours sincerely,

for Professor John Pearn
Chair
Queensland Children's Health Services (RCH) Human Research Ethics Committee

Cc: Ethics Committee Files

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CHILDREN'S HEALTH SERVICES QUEENSLAND
HUMAN RESEARCH ETHICS COMMITTEE

Professor John Pearn (Chair) 3365 5323
Mrs Amanda Smith (Co-ordinator) 3636 9167

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Herston QLD 4029 Australia
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1st November 2012

Ms Katherine Benfer
Speech Pathologist
QCPRRC
Level 7, Block 6
Royal Women's & Brisbane Hospital
Herston QLD 4029

Dear Ms Benfer,

HREC Reference number: HREC/08/QRCH/112
Project title: The Queensland Cerebral Palsy Child Study - Growth, Nutrition And Physical Activity.
Amendment number: HREC/08/QRCH/112/AM07

Many thanks for your letter of the 20th September regarding an amendment to the above project. This has now been reviewed and I am pleased to advise that the amended documents reviewed and approved were:

Document	Version	Date
Notification of amendment		
Covering Letter		20 September 2012

The QLD Children's Health Services (RCH) Human Research Ethics Committee (HREC) is constituted and operates in accordance with the National Health and Medical Research Council's *"National Statement on Ethical Conduct in Human Research (2007), NHMRC and Universities Australia Australian Code for the Responsible Conduct of Research (2007)* and the *"CPMP/ICH Note for Guidance on Good Clinical Practice"*.

A copy of this letter must be forwarded to the Research Governance Officer. It should be noted that all requirements of the original approval still apply.

Yours sincerely,

Professor John Pearn
Chair
Children's Health Services Queensland Human Research Ethics Committee

Cc: Ethics Committee Files

CHILDREN'S HEALTH SERVICES QUEENSLAND
HUMAN RESEARCH ETHICS COMMITTEE

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Herston QLD 4029 Australia
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11th February 2013

Ms Katherine Benfer
NHMRC PhD Scholar/Speech Pathologist
Queensland Cerebral Palsy & Rehabilitation Research Centre
Level 7, Block 6
Royal Brisbane and Women's Hospital
Herston, QLD 4029

Dear Ms Benfer,

HREC Reference number: HREC/08/QRCH/112
Project title: The Queensland Cerebral Palsy Child Study - Growth, Nutrition and Physical Activity.
Amendment number: HREC/08/QRCH/112/AM09

Many thanks for your letter of the 4th February regarding an amendment to the above study. This has now been reviewed and I am pleased to advise that the amended documents reviewed and approved were:

Document	Version	Date
Covering Letter		04 February 2013
Parent/Guardian Information Sheet and Consent Form (Form E)	3	04 February 2013

The QLD Children's Health Services (RCH) Human Research Ethics Committee (HREC) is constituted and operates in accordance with the National Health and Medical Research Council's *"National Statement on Ethical Conduct in Human Research (2007), NHMRC and Universities Australia Australian Code for the Responsible Conduct of Research (2007)* and the *"CPMP/ICH Note for Guidance on Good Clinical Practice".*

A copy of this letter must be forwarded to the Research Governance Officer. It should be noted that all requirements of the original approval still apply.

Yours sincerely,

for

Professor Alan Isles
Deputy Chair
Children's Health Services Queensland Human Research Ethics Committee

Cc: Ethics Committee Files

CHILDREN'S HEALTH SERVICES QUEENSLAND
HUMAN RESEARCH ETHICS COMMITTEE

Professor John Pearn (Chair) 3365 5323
Mrs Amanda Smith (Co-ordinator) 3636 9167

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Level 3, RCH Foundation Building
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8th May 2013

Ms Katherine Benfer
Queensland Cerebral Palsy and Rehabilitation Research Centre
The University of Queensland
Level 7, Block 6
Royal Brisbane and Women's Hospital
Hertson, QLD 4029

Dear Ms Benfer,

HREC Reference number: HREC/13/QRCH/69
Project title: Queensland Cerebral Palsy Child: Growth, Nutrition and Physical Activity -- Oropharyngeal Dysphagia in Bangladesh

Many thanks for your letter of the 1st May with responses to queries raised by the Committee in relation to the above project. This has now been reviewed.

This HREC is constituted and operates in accordance with the National Health and Medical Research Council's (NHMRC) *National Statement on Ethical Conduct in Human Research (2007), NHMRC and Universities Australia Australian Code for the Responsible Conduct of Research (2007)* and the *CPMP/ICH Note for Guidance on Good Clinical Practice.*

I am pleased to advise the proposal meets the requirements of the National Statement on Ethical Conduct in Human Research and the Committee is happy to give approval.

This project has Ethics approval for the following sites:

- Royal Children's Hospital, Brisbane

[Note: If additional sites are engaged prior to the commencement of, or during the research project, the Coordinating Principal Investigator is required to notify QLD Children's Health Services (RCH) Human Research Ethics Committee (HREC). Notification of withdrawn sites should also be provided to the QLD Children's Health Services (RCH) Human Research Ethics Committee (HREC) in a timely fashion.

The documents reviewed and approved include:

Document	*Version*	*Date*
Investigator CV	1	15 April 2013
Letter from Sponsor	1	12 March 2013
Protocol	1	15 April 2013
Study flyer	1	15 April 2013
Research Funding Schedule	1	15 April 2013
Questionnaire: Feeding evaluation forms (SOMA, DDS, PSAS)	1	15 April 2013

Parent child interaction behaviour assessment (Moore)	1	15 April 2013
Questionnaire: Feeding Questionnaire: OPD-Bd	1	15 April 2013
Physician's Checklist: OPD-Bd	1	15 April 2013
USDA Food Security Survey Module	1	15 April 2013
Poverty Measurement Tool	1	15 April 2013
Multidimensional Scale of Perceived Social Support	1	15 April 2013
Anthropometry and Impedance	1	15 April 2013
Weighed food diary record form	1	15 April 2013
Health Resource Use Form	1	15 April 2013
Application		
Covering Letter		01 May 2013
Study Flyer		
Participant Video and Photographic Consent	2	01 May 2013
Parent/Guardian Information Statement & Consent Form	2	01 May 2013
Response to Request for Further Information		

Please note the following conditions of approval:

1. We require an annual progress report (or sooner if the project is completed) concerning the study. This must include progress to date or outcome in the case of completed research. (In accordance with National Statement 5.5.3)

2. HREC approval is valid from 8/5/13 – 8/5/16.

3. In accordance with the National Statement (3.3.12), before beginning the clinical phase of the research, researchers should register clinical trials in a publicly accessible domain.

4. If the project does not proceed, the Committee must be informed as soon as possible. (In accordance with National Statement 5.5.6)

5. The Committee must be informed of any potential or realised problem with bioethical implications, if such occurs during the conduct of the research project.

6. Any serious adverse event (SAE) that arises in the context of this research, or involving a researcher conducting this research, must be reported to the Ethics Committee within 72 hours and reported to the sponsor (if applicable) within the stipulated time frame.

   Serious Adverse Event Reports that are generated off-site may be (a) Serious Unexpected Adverse Reactions or (b) Serious Events which the Research Team believes cannot be related to the research intervention. The Research team must report incidents of (a) during multi-centre trials. Such are required to be submitted to the Chair of HREC on receipt by the researcher. A summary of the SAE reports is to accompany the submission. Information required includes; patient details (age & sex), adverse event, outcome and the likelihood of the event being related to the study drug/device/procedure.

   With respect to all SAEs, the researcher must provide his or her opinion as to whether the SAE is directly related to the research intervention. A copy of the SAE Summary must be provided. (This can be obtained from the Ethics Officer)

7. Amendments to the research project which may affect the ongoing ethical acceptability of a project must be submitted to the HREC for review. Major amendments should be reflected in a revised online NEAF (accompanied by all relevant updated documentation and a cover letter from the principal investigator, providing a brief description of the changes, the rationale for the changes, and their implications for the ongoing conduct of the study). Hard copies of the revised NEAF, the cover letter and all relevant updated documents with tracked changes must also be submitted to the HREC and the RGO as per standard HREC/RGO SOP. Further advice on submitting amendments is available from: http://www.health.qld.gov.au/ohmr/documents/regu/resrch_user_guide_v1.pdf

8. The Ethics Committee may conduct a randomly identified audit of a proportion of research projects approved by the Committee. That audit process will look at such issues as;
   a. Security of Documents
   b. Consent Form Register
   c. Serious Adverse Events Register
   d. Withdrawal of Participants – who and why
   e. The de-identification of data

9. Ethical approval to undertake this research project is given on the understanding that you have an intention to publish your findings in a refereed journal or similar peer-reviewed forum. If you do not have this intention, it is an absolute requirement that you notify the Ethics Committee formally. In this latter instance, approval for this research is not given at this time; and will require further negotiation. Your work must be in accordance with the following:

   - National Statement on Ethical Conduct in Human Research: ***http://www.nhmrc.gov.au/guidelines/publications/e72-0***
   - Queensland Health Management Research Policy: ***http://www.health.qld.gov.au/ohmr/html/regu/resrch_mge_policy.asp***
   - Declaration of Helsinki: ***http://www.wma.net/en/30publications/10policies/b3/17c.pdf***
   - Guidelines under Section 95 of the Privacy Act1995 and Guidelines approved under Section 95A of the Privacy Act 1995. ***http://www.health.qld.gov.au/ohmr/html/regu/aces_conf_hth_info.asp***
   - Queensland Health Privacy Guidelines IS42 & IS42A: ***http://www.health.qld.gov.au/privacy/IS42A.asp***

10. Researchers should note, if not QLD Health employees, a Blue Card may be required for contact with children.

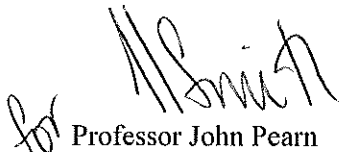
11. The Researcher must send the **'Notification of Commencement of Research Protocol'** as soon as research begins. Status of the project will remain as 'Not Started' until this form is received.

Should you have any queries about the HREC's consideration of your project please contact Amanda Smith (Co-ordinator) or Professor John Pearn (Chairperson). The HREC terms of Reference, Standard Operating Procedures, membership and standard forms are available from: http://www.health.qld.gov.au/ohmr/html/regu/regu_home.asp

You are reminded that this letter constitutes ethical approval only. This project cannot proceed at any site until separate research governance authorisation has been obtained from the CEO or Delegate of the institution under whose auspices the research will be conducted at that site.

The HREC wishes you every success in your research.

Yours sincerely,



Professor John Pearn
Chair
QLD Children's Health Services (RCH) Human Research Ethics Committee

Cc: Ethics Committee Files

THE UNIVERSITY OF QUEENSLAND

Institutional Human Research Ethics Approval

Project Title:	Queensland Cerebral Palsy Child: Growth, Nutrition And Physical Activity - Oropharyngeal Dysphagia In Bangladesh Substudy
Chief Investigator:	Ms Katherine Benfer, Prof Roslyn Boyd, Ms Kelly Weir, Prof Peter Davies, Dr Kristie Bell, Dr Robert Ware, Mr Jahangir Alam, Dr Baitun Nahar
Supervisor:	Prof Roslyn Boyd, Ms Kelly Weir, Prof Peter Davies
Co-Investigator(s):	Dr Sabera Bilkis, Ms Hosneara Perveen, Ms Fatema Akhter Mitu, Dr Sasaka Bandaranyake, Ms Laura Pareezer, Ms Rachel Jordan, Ms Christine Finn, Prof Tahmeed Ahmed
School(s):	School of Medicine
Approval Number:	2013000625
Granting Agency/Degree:	NHMRC; Graduate Student International Travel Award
Duration:	1st July 2014

Comments:

Expedited review on the basis of approval from the Queensland Children's Health Services HREC (RCH) dated 08/05/2013

Note: If this approval is for amendments to an already approved protocol for which a UQ Clinical Trials Protection/Insurance Form was originally submitted, then the researchers must directly notify the UQ Insurance Office of any changes to that Form and Participant Information Sheets & Consent Forms as a result of the amendments, before action.

Name of responsible Committee:
Medical Research Ethics Committee
This project complies with the provisions contained in the *National Statement on Ethical Conduct in Human Research* and complies with the regulations governing experimentation on humans.

Name of Ethics Committee representative:
Professor Bill Vicenzino
Chairperson
Medical Research Ethics Committee

Signature _____ Date 24/5/13

পক্ষাঘাতগ্রস্তদের পুনর্বাসন কেন্দ্র (সিআরপি)
Centre for the Rehabilitation of the Paralysed (CRP)
a project of the Trust for the Rehabilitation of the Paralysed
Head Office: CRP-Savar, CRP-Chapain, Savar, Dhaka-1343, Bangladesh
Tel: +880 (0)2-7745464-5, Fax: 7745069, E-mail: contact@crp-bangladesh.org, Website: www.crp-bangladesh.org

Ref: CRP/RE/0401/55 Date: 11/08/2013

Re: Ethics Approvals for Queensland Cerebral Palsy Child: Growth, Nutrition and Physical Activity – Oropharyngeal Dysphagia in Bangladesh Sub study

Chief Investigator: Ms Katherine Benfer, Prof Roslyn Boyd, Ms Kelly Weir, Prof Peter Davies, Dr Kristie Bell, Dr Robert Ware, Mr Jahangir Alam, Dr Baitun Nahar

Co-Investigator: Dr Sabera Bilkis, Ms Hosneara Perveen, Ms Fatema Akhter Mitu, Dr Sasaka Bandaranyake, Ms Laura Pareezer, Ms Rachel Jordan, Ms Christine Finn, Prof Tahmeed Ahmed

We understand that this study has gained the following ethics approvals:

- Queensland Children's Health Services Human Research Ethics Committee
- University of Queensland Medical Research Ethics Committee
- Centre for the Rehabilitation of the Paralysed Ethics Review Committee
- International Centre for Diarrhoeal Disease Research, Bangladesh (icddr,b) Ethics Review Committee (pending)

We approve the research to be conducted within the Centre for the Rehabilitation of the Paralysed with the fore-mentioned ethics approvals.

Md Shafiq-ul Islam
Executive Director
Centre for the Rehabilitation of the Paralysed (CRP)

CENTRE FOR THE REHABILITATION OF THE PARALYSED
Chapain
Po-CRP Chapain
Savar, Dhaka.

icddr,b

Memorandum

13 June 2013

To: Dr.Baitun Nahar
Principal Investigator of research protocol # PR-13047
Centre for Nutrition and Food Security(CNFS)

From: Abbas Bhuiya, Ph.D.
Chairperson
Research Review Committee

Sub: Approval of research protocol # PR-13047

Thank you for your memo dated 11 June 2013 requesting for approval of your research protocol # PR-13047 titled "**Queensland Cerebral Palsy Child: Growth, Nutrition and Physical Activity – Oropharyngeal Dysphagia in Bangladesh**" waiving normal RRC review process since the protocol was reviewed and approved by QLD Children's Health Services (RCH) Human Research Ethics Committee. I have the pleasure to approve the protocol and you are advised to proceed the implementation of the research protocol subject to the approval by the Ethical Review Committee (ERC).

Terms of approval

1. The research protocol is approved for 12-month period from the date of approval of the protocol by the Ethical Review Committee. Approval for further continuation of the research work, if needed, shall be obtained before expiration of the initial approval.

2. You should notify the IRB Secretariat of the start date of the protocol for updating in the integrated navision system. The protocol start date will not be updated in the navision system until receiving information from you. Therefore you will not be able to operate budget code and continue spending funds under the research protocol.

3. The RRC approval shall automatically be revoked after one year if the protocol is not started. After one year, you shall have to seek approval for revalidation of the protocol by the RRC & ERC before starting the protocol.

4. This approval is only valid whilst you hold a position at icddr,b; and in the event of your departure from the Centre, a new Principal Investigator will be designated for the research protocol.

GPO Box 128, Dhaka 1000, Bangladesh.
For Packages : 68, Shaheed Tajuddin Ahmed Sarani, Mohakhali, Dhaka 1212, Bangladesh.
Phone : 880-2-8860523-32, Web : http://www.icddrb.org, Fax : 880-2-8823116.

5. You should notify the RRC and the ERC immediately of any serious or unexpected adverse effects on participants or unforeseen events that might affect continued acceptability of the protocol.

6. Any changes to the research protocol require the submission (in prescribed form) and approval of an amendment/addendum. Substantial variations may require a new protocol.

7. Continued approval of this protocol is dependent on your periodically updating the Centre's database for the protocol to show the progress; and a final report/completion report, including data set, must be submitted to the IRB secretariat for consideration by the RRC and ERC at the conclusion of the protocol within three months of its completion.

8. You shall submit a report for time extension of the protocol (in prescribed form) if you are unable to complete the protocol activities within the time mentioned in the protocol.

9. You are responsible for systematic storage and retention of the original data pertaining to the research protocol; and the ownership of data after certain period shall be determined as per Centre's rules and regulations.

10. The RRC should be notified if the protocol is discontinued before the expected date of completion.

I wish you all the success in conducting the research protocol.

Thank you.

Cc: Director, CNFS

icddr,b

Memorandum

19 August 2013

To: Dr Baitun Nahar
Principal Investigator of research protocol # PR-13047
Centre for Nutrition and Food Security (CNFS)

From: Professor K Z Mamun
Chairperson
Ethical Review Committee (ERC)

Sub: Approval of research protocol # PR-13047

Thank you for your memo dated 17 August 2013 attaching the modified version of your research protocol # PR-13047 titled **"Oropharyngeal dysphagia and its relationship to dietary intake, nutritional status and gross motor skills in children with cerebral palsy in Bangladesh"** addressing the issues raised by the committee in its special meeting held on 7 July 2013 to the satisfaction of the Committee. Accordingly, the Committee approved the research protocol. You will be required to observe the following terms and conditions in implementing the research protocol:

1. The research protocol is approved for 12-month period from the date of approval of the protocol by the Ethical Review Committee. Approval for further continuation of the research work, if needed, shall be obtained before expiration of the initial approval.

2. The ERC approval shall automatically be revoked after one year if the protocol is not started. After one year, you shall have to seek approval for revalidation of the protocol by the RRC & ERC before starting.

3. You should notify the IRB Secretariat of the start date of the protocol for updating in the integrated navision system. The protocol start date will not be updated in the navision system until receiving information from you. Therefore you will not be able to operate budget code and continue spending funds under the research protocol.

4. As Principal Investigator, the ultimate responsibility for scientific and ethical conduct including the protection of the rights and welfare of study participants vest upon you. You shall also be responsible for ensuring competence, integrity and ethical conduct of other investigators and staff directly involved in this research protocol.

5. You shall conduct the study in accordance with the ERC-approved protocol and shall fully comply with any subsequent determinations by the ERC.

6. You shall obtain prior approval from the Research Review Committee and the ERC for any modification in the approved research protocol and/or approved consent form(s), except in case of emergency to safeguard/eliminate apparent immediate hazards to study participants. Such changes must immediately be reported to the ERC Chairman.

7. You shall recruit/enrol participants for this study strictly adhering to the criteria mentioned in the research protocol.

8. You shall obtain legally effective informed consent (i.e. consent should be free from coercion or undue influence) from the selected study participants or their legally responsible representative, as approved in the protocol, using the approved consent form prior to their enrollment in this study. Before obtaining consent, all prospective study participants must be adequately informed about the purpose(s) of the study, its methods and procedures, and also what would be done if they agree and also if they do not agree to participate in the study.

   They must be informed that their participation in the study is voluntary and that they can withdraw their participation any time without any prejudice. Signed consent forms should be preserved for a period of at least five years following official termination of the study.

9. You shall promptly report the occurrence of any Serious Adverse Event or unanticipated problems of potential risk to study participants or others to the ERC in writing within 24 hours of such occurrences.

10. Any significant new findings, developing during the course of this study that might affect the risks and benefits and thus influence either participation in the study or continuation of participation should be reported in writing to the participants and the ERC.

11. Data and/or samples should be collected and interviews should be conducted, as specified in the ERC-approved protocol, and confidentiality must be maintained. Data/samples must be protected by reasonable security, safeguarding against risks such as their loss or unauthorized access, destructions, used by others, and modification or disclosure of data. Data/samples should not be disclosed, made available to or use for purposes other than those specified in the protocol, and shall be preserved for a period, as specified under Centre's policies/practices.

12. You shall promptly and fully comply with the decision of the ERC to suspend or withdraw its approval for the research protocol.

13. You shall report progress of research to the ERC for continuing review of the implementation of the research protocol as stipulated in the ERC Guidelines. Relevant excerpt of ERC Guidelines and '*Annual/Completion Report for Research Protocol involving Human Subjects*' are attached for your information and guidance.

14. The RRC should be immediately notified if the protocol is discontinued before the expected date of completion.

I wish you success in running the above-mentioned study.

Cc: Director, CNFS

CHILDREN'S HEALTH SERVICES QUEENSLAND
HUMAN RESEARCH ETHICS COMMITTEE

Professor John Pearn (Chair) 3365 5323
Mrs Amanda Smith (Co-ordinator) 3636 9167

Queensland Government

Level 3, RCH Foundation Building
Royal Children's Hospital
Herston QLD 4029 Australia
Telephone (07) 3636 9167
Facsimile (07) 3365 5455

26th November 2013

Ms Katherine Benfer
Queensland Cerebral Palsy and Rehabilitation Research Centre
The University of Queensland
Level 7, Block 6
Royal Brisbane and Women's Hospital
Herston, QLD 4029

Dear Ms Benfer,

HREC Reference number: HREC/13/QRCH/69
Project title: Queensland Cerebral Palsy Child: Growth, Nutrition and Physical Activity -- Oropharyngeal Dysphagia in Bangladesh
Amendment number: HREC/13/QRCH/69/AM01

Many thanks for your letter of the 22nd November regarding an amendment to the data storage location for the above project. This has now been reviewed and the Committee is happy to give approval for this change.

The QLD Children's Health Services (RCH) Human Research Ethics Committee (HREC) is constituted and operates in accordance with the National Health and Medical Research Council's *"National Statement on Ethical Conduct in Human Research (2007), NHMRC and Universities Australia Australian Code for the Responsible Conduct of Research (2007)* and the *"CPMP/ICH Note for Guidance on Good Clinical Practice"*.

A copy of this letter must be forwarded to the Research Governance Officer. It should be noted that all requirements of the original approval still apply.

Yours sincerely,

For Professor John Pearn
Chair
Children's Health Services Queensland Human Research Ethics Committee

Cc: Ethics Committee Files

THE UNIVERSITY OF QUEENSLAND

Institutional Human Research Ethics Approval

Project Title:	Queensland Cerebral Palsy Child: Growth, Nutrition And Physical Activity - Oropharyngeal Dysphagia In Bangladesh Substudy - 28/11/2013 - AMENDMENT
Chief Investigator:	Ms Katherine Benfer, Prof Roslyn Boyd, Ms Kelly Weir, Prof Peter Davies, Dr Kristie Bell, Dr Robert Ware, Mr Jahangir Alam, Dr Baitun Nahar
Supervisor:	Prof Roslyn Boyd, Ms Kelly Weir, Prof Peter Davies
Co-Investigator(s):	Dr Savera Bilkis, Ms Hosneara Perveen, Ms Fatema Akhter Mitu, Dr Sasaka Bandaranyake, Ms Laura Pareezer, Ms Rachel Jordan, Ms Christine Finn, Prof Tahmeed Ahmed
School(s):	School of Medicine
Approval Number:	2013000625
Granting Agency/Degree:	NHMRC; Graduate Student International Travel Award
Duration:	1st July 2014

Comments/Conditions:

Note: if this approval is for amendments to an already approved protocol for which a UQ Clinical Trials Protection/Insurance Form was originally submitted, then the researchers must directly notify the UQ Insurance Office of any changes to that Form and Participant Information Sheets & Consent Forms as a result of the amendments, before action.

Name of responsible Committee:
Medical Research Ethics Committee
This project complies with the provisions contained in the *National Statement on Ethical Conduct in Human Research* and complies with the regulations governing experimentation on humans.

Name of Ethics Committee representative:
Professor Bill Vicenzino
Chairperson
Medical Research Ethics Committee

Signature _____ Date 4 Dec 2013

Queensland cerebral palsy & rehabilitation research centre

Queensland Government
Queensland Health

Children's Health Service District - Royal Children's Hospital
PARENT/GUARDIAN INFORMATION STATEMENT
AND CONSENT FORM

Research Project Title: Queensland Cerebral Palsy Child Study: Nutrition, Growth and Physical Activity of Children

Researchers: A/Prof Peter Davies, A/Prof Roslyn Boyd, Dr Kristie Bell, Dr Sean Tweedy, Prof Richard Stevenson, Dr Stewart Trost, Dr Robert Ware, Ms Kelly Weir, Dr Lynne McKinlay, Dr Kate Sinclair, Paula Luck, Fiona Caristo, Jacqueline Walker and Laura Pareezer.

Thank you for taking the time to read this information Statement. This Information Statement and Consent Form is 7 pages long. Please make sure you read all pages.
For people who speak languages other English: If you would also like information about the research and a Consent Form in your language, please ask the person explaining this project to you.

You are invited to participate in the research project that is explained below.
What is an Information Statement?
These pages tell you about the research project. It explains to you all the steps and procedures of the project. The information is to help you decide whether or not you would like your child to take part in the research. Please read this Information Statement carefully. You are welcome to ask us questions about anything in it. You may wish to talk about the project with your family, friends or health care worker.

Participation in this research is entirely voluntary. If you don't want your child to take part, you don't have to. You can withdraw your child from the study at any time without explanation and there will be no penalty from any staff at the Royal Children's Hospital or the University of Queensland. Withdrawal will not affect your child's care in any way.

What is this research project about?
This project is about growth, nutrition, diet and physical activity of children who have cerebral palsy. Cerebral palsy is a physical disability caused by early brain injury. It occurs in 1 in 500 children. Children with cerebral palsy may be shorter and thinner than their typically developing peers. This project will look at how eating and drinking skills, dietary intake, and the amount of physical activity that children with cerebral palsy do effects the way they grow and develop, their quality of life, participation and the amount of health care used.

Children will attend specialist "QLDCPchild: Growth, Nutrition and Physical Activity clinics" at the Royal Children's Hospital in Brisbane or at one of the outreach clinics of the Queensland Paediatric Rehabilitation Service (QPRS) or the Queensland Cerebral Palsy Health Service (CP Health) three times between the age of 18 months and 5 years (see later for details). These assessments will be organised to coincide with appointments you may already have to minimise any additional visits to the RCH.

Who are the researchers?

- Associate Professor Peter Davies is the Director of the Children's Nutrition Research Centre at the University of Queensland.
- Associate Professor Roslyn Boyd is a Paediatric Physiotherapist and Scientific Director at the Queensland Cerebral Palsy and Rehabilitation Research Centre, University of Queensland and the Royal Children's Hospital (Brisbane).
- Dr Kristie Bell is a Paediatric Dietician at the Royal Children's Hospital and the University of Queensland. She will coordinate the project and supervise the assessments.
- Professor Richard Stevenson is a Paediatrician at the Kluge Children's Rehabilitation Centre in the United States of America.
- Dr Sean Tweedy is an Exercise Physiologist at the University of Queensland.
- Ms Kelly Weir is a speech pathologist at the University of Queensland and the Royal Children's Hospital, she will analyse the video of your child's eating.
- Dr Lynne McKinlay is a Rehabilitation Specialist and Director of the Department of Rehabilitation at the Royal Childrens Hospital. She will discuss the the diagnosis of cerebral palsy.
- Dr Kate Sinclair is a neurologist at the Royal Children's Hosptial. She will discuss the diagnosis of cerebral palsy.
- Associate Professor Stewart Trost from the Department of Nutrition and Exercise Science, Oregon State University, will provide advice regarding the collection of the physical activity data.
- Dr Robert Ware is a statistician with the University of Queensland.

Why is my child being asked to be in this research project?

We are asking your child to take part because he/she has delayed motor development that may be due to cerebral palsy and was born in Queensland in one of the following years: 2006, 2007, 2008 or 2009.

What are the alternatives to taking part in this project?

There is no obligation to participate in this project. Should you choose not to participate in this project, your child will have all the usual access to treatment at the Royal Children's Hospital and District Health Service.

What does my child need to do to be in this research project?

Your child will be seen 3 times at:

1. between 18 - 30 months,
2. between 36 - 42 months
3. 5 years of age.

Each visit will take approximately 1.5 to 2 hours in total. At each of these visits the following assessments will be performed:-

1. Classification of motor type, distribution and severity of cerebral palsy by the research physiotherapist.
2. Medical review: a medical professional will (a) confirm the diagnosis of cerebral palsy by identifying the early medical history; (b) review your child's medical status; (c) may order a pelvic radiograph to monitor hip displacement (if required) and (d) will review whether your child has had a brain MRI and order an MRI for confirmation of diagnosis (if required). MRI to occur after 24 months of age.

3. Anthropometry

a. Growth:- We will measure your child's height or length and weight as well as their knee height, upper arm length, head circumference and upper arm circumference.

b. Skinfold Thickness:- The thickness of the skin will be measured at two sites: one on the back of the upper arm (tricep) and one under the shoulder blade (subscapular). This will provide information regarding your child's body fat stores.

4. Body Composition:- The following two methods will provide information regarding the amount of water in your child's body.

a. Bioelectrical impedance analysis:- This is a simple, painless and safe technique to measure body composition. The technique requires that your child lie quietly for a few minutes with surface electrodes taped lightly to their wrist and ankle. A very small electrical current passes through the body. This current is completely safe and so mild that it cannot be felt. The measurement only takes a few seconds during which your child will not be able to wear shoes, socks and metallic jewelry.

b. Heavy water:- This is a very simple technique that involves your child drinking a special type of water called deuterium that we then measure the concentration of in their urine. Deuterium is naturally occurring, non-toxic and non-radioactive and tastes exactly the same as tap water. It is totally harmless and has been used in worldwide studies from premature babies to pregnant women and the elderly. All you need do is collect a single urine sample prior to the dose and a second one 5 hours after. The urine samples will be collected from you by a certified courier at a time that is convenient to you.

3. Feeding Evaluation:- You will be asked to bring a small snack to the hospital for your child to consume during your visit. The type of snack will be discussed with you prior to the visit. Your child will be videotaped whilst eating this snack. The video will be reviewed by a speech pathologist to determine if your child has any difficulty with eating.

4. Physical Activity:- Following your appointment your child will be required to wear a small activity monitor called an Actigraph around their waist. Your child needs to wear the Actigraph every day for 3 days whilst they are awake. It can be taken off when your child goes to bed and put back on when they wake. It can also be taken off when your child bathes or goes swimming. You will be asked to record the time of day when the monitor is worn on a form provided.

5. Dietary Intake:- Following your appointment you will be required to record all food and drink consumed by your child over a 3 day period on a form provided. Detailed instructions regarding how to do this will be discussed with you and provided on a separate form.

6. Questionnaires:- You will be asked to complete questionnaires regarding the following:
    a. Your child's feeding ability and eating/drinking skills
    b. Participation: using the Paediatric Evaluation of Disability Inventory (PEDI)
    c. Quality of life: using the parend-report condition specific measure (CP-QOL-child)
    d. A record of what treatments and interventions your child receives

Magnetic Resonance Image Scan (MRI) of your child's brain

If your child has not had a brain Magnetic Resonance Imaging (MRI) Scan previously, it will be offered after your child turns 24 months old. The scanner will take pictures of your child's brain using magnetic and radio waves. No X- rays are used. Your child will have an anaesthetic for the MRI scan as it is a very noisy and constrained environment that may be frightening, and the child also needs to be completely still for the test. MRI brain scans are routinely done at this age for infants who have a suspected brain injury to determine the nature of the brain injury. If you would prefer your child not to have an anaesthetic, we can attempt to perform the scan without an

anaesthetic while your child is sleeping. To prepare your child for this, we will give you a tape recording of the scanner sounds that you can play to your child at home in the evenings around the time that your child is going to sleep. The child will then become accustomed to the unusual noises that the scanner makes. We will then do the scan at the Royal Children's Hospital at a time when your child would be asleep (evening). Your child will be positioned on a comfortable pillow in the scanner and monitored over the scan time (approximately 30 – 60 minutes). The risks associated with performing an anesthetic are the same as they are for any anaesthetic, there is no additional risk for it being performed in the MRI. The formal report of the scan will be given to you along with a time to discuss the results with a member of our research team. Although the MRI is offered and may provide helpful information, your child can participate in the study even if you choose not to have the MRI scan.

The MRI visit is an additional visit to the hospital. If you are traveling from outside Brisbane we will pay for the costs of travel and parking, both for the scan and the other visits.

How will this study benefit my child?

The study will provide you with information about your child's growth and dietary intake. You will have the opportunity to discuss your child's progress and any concerns with the research team. The final study results will be summarized and reported back to you at the conclusion of the study. You will have the opportunity to have an MRI of your child's brain. This may be helpful for providing advice about the cause of the cerebral palsy and any associated genetic implications (which are unusual). You will have the opportunity to discuss your child's progress and any concerns with the research team.

How will this study benefit other people in the future?

The results of this study will provide valuable information that will help us to identify why some children with cerebral palsy grow poorly and how poor growth, dietary intake and physical activity may impact on their quality of life, participation and the amount of health care used. It will also assist us to determine which children need help to improve their nutrition, growth and physical activity and at what age is the best time to do this. In addition, it will provide us with information about how well different methods can measure the body composition of children with cerebral palsy and allow us to make recommendations on their use for others working with children with cerebral palsy.

What are the risks for my child?

There are no additional risks for your child with these measurements (including Magnetic Resonance Imaging (MRI)) over and above that experienced in every day life. All procedures are safe and are frequently used for clinical and research purposes. MRI scans are routinely performed under general anaesthesia for many children with cerebral palsy. You are under no obligation to consent to your child having a brain MRI scan.

What happens if something abnormal or unexpected is found in my child's MRI scan?

In this study, we will take a number of pictures of your child's brain, or will review pictures that have already been taken. After your child's scan, a specialist will examine these pictures. This will not be done on the day of the scan. With cerebral palsy there is a high chance of finding an abnormality on the brain scan. There is the possibility that the scan will show up something in your child's brain that we had not expected. If this happens, we will arrange for you to meet with a medical professional who can explain the findings to you. If any of the results of the scan are distressing for you, we will offer you counseling with specially trained staff.

What are the possible discomforts and/or inconveniences?
The MRI scanner is noisy so that protective earmuffs will be placed over your child's ears during the scan. The only inconvenience to you and your child is the time that the assessments will take, and the trips you will have to make to the hospital.

The assessment appointments will be planned to minimize any inconvenience to you and to coincide with any other appointment that you may have at the hospital. The assessments will take about 2 to 2.5 hours in total and you will be required to come to the hospital on 3 occasions over a 3 year period. We will pay for the cost of parking your car at the hospital during these visits. In addition, once home you will be required to complete a 3 day food diary and your child will need to wear a physical activity monitor for 3 days.

What will be done to make sure the information is confidential?
Data from these assessments will be stored electronically without your child's name. A number will be used to identify them. This number will be linked to your child's name and the linking file will be kept confidential and only made available to the researchers. A separate database will contain your contact information and those results required for the generation of clinical reports. All databases will be password protected with limited access available to the researchers involved in the study.

Data collection sheets recording the assessments and the videotapes of the assessments will be stored in an individual file for your child in a secure, locked, fire proof filing cabinet. Only the researchers will have access to this information. These data sheets will be kept for 7 years at the Royal Children's Hospital. If we give talks or write about the results of this project, we will not use any names.

All names and identifying information will be removed from data prior to any analysis.

Will I be informed of the results when the research project is finished?
You will receive a written report about your child's progress after each visit. If at any time you would like more information about your child's results, an appointment will be organized with one of the researchers. A regular newsletter will also be sent to you about the progress of the study. At the end of the study all families will be sent a summary of the results. The newsletters and final summary will talk about the children as a group and your child will not be identified in person.

You can decide whether or not to give permission for your child to take part in this research project. You can decide whether or not you would like to withdraw your child at any time without explanation. Your decision whether or not for your child to participate will not prejudice your child's future relations with the Royal Children's Hospital and District Health Service. If you decide for your child to participate, you are free to withdraw your consent and discontinue participation at any time. The decision to withdraw from the study will not affect their routine medical treatment or their relationship with the people treating them. You may like to discuss your child's participation in this research project with your family and with your doctor. You can ask for further information before deciding to take part.

If you would like more information about the study or if you need to contact a study representative in an emergency, the person to contact is:

Name: A/Prof Roslyn Boyd, A/Professor Cerebral Palsy and Rehabilitation Research,
Contact telephone: (07) 3365 5327 **mobile:-** 0434 608 443

Or

Name: Dr Kristie Bell, Clinical Postdoctoral Research Fellow
Contact telephone: (07) 3636 5536.

What are my child's rights as a participant?

I am informed that except where stated above, no information regarding my child's medical history will be released. This is subject to legal requirements. I am informed that the results of any tests involving my child will not be published so as to reveal my child's identity. This is subject to legal requirements. The detail of the procedure proposed has also been explained to me. This includes how long it will take, how often the procedure will be performed and whether any discomfort will result. It has also been explained that my child's involvement in the research may not be of any benefit to him or her. I understand that the purpose of this research project is to improve the quality of medical care in the future. I have been asked if I would like to have a family member or a friend with me while the project is explained to me. I understand that this project follows the guidelines of the National Statement on Ethical Conduct in Research Involving Humans (1999).I understand that this research project has been approved by the Royal Children's Hospital Ethics Committee on behalf of the Royal Children's Hospital and Health Services District, Brisbane. I have received a copy of this document.

Contact:-

The Research Ethics Committee of the Royal Children's Hospital and Health Services District has approved this study. Should you wish to discuss the study with someone not directly involved, in particular in relation to matters concerning policies, information about the conduct of the study or your rights as a participant, or you wish to make a confidential complaint, at any time, you may contact the Co-ordinator on the ethics committee, Royal Childrens Hospital and Health Services District, c/o Dept of Pediatrics and Child Health, Level 3, Foundation building, Herston. QLD. 4029. This study adheres to the Guidelines of the ethical review process of The University of Queensland. Whilst you are free to discuss your participation in this study with project staff (contactable on 07 3636 5542), if you would like to speak to an officer of the University not involved in the study, you may contact the Ethics Officer on 3365 3924.

This study has been approved by the Cerebral Palsy League of Queensland Ethics Committee (**CPLQ- 2009/10 – 1029**). If needed, verification can be obtained by writing or telephoning the Cerebral Palsy League Ethics Committee, c/-Cerebral Palsy League of Queensland, 55 Oxlade Drive, New Farm, Brisbane Qld 4005 or PO Box 386, Fortitude Valley QLD 4006 Tel: 07 33588101

Queensland cerebral palsy & rehabilitation research centre

Queensland Government
Queensland Health

STANDARD INFORMED CONSENT FOR PARENT/GUARDIAN TO GIVE CONSENT FOR THEIR CHILD TO PARTICIPATE IN A RESEARCH PROJECT

Project Number

EHRC No 2008002260

Title of Project

QLD CP Child study: Nutrition, Growth and Physical Activity

Investigator(s)

Prof Peter Davies, A/Prof Roslyn Boyd, Dr Kristie Bell, Dr Sean Tweedy, Prof Richard Stevenson, Dr Stewart Trost, Dr Robert Ware, Ms Kelly Weir, Dr Lynne McKinlay, Dr Kate Sinclair, Paula Luck, Fiona Caristo, Jacqueline Walker and Laura Pareezer.

I (Parent/Guardian name) _____

voluntarily consent for my child to take part in the above titled Research Project, explained to me by

Mr/Ms/Dr/Professor _____

Child's Name _____

Address _____

Contact Phone Numbers _____

I (Parent/Guardian name) _____

voluntarily consent for my child to take part in the above titled Research Project, explained to me by

Mr/Ms/Dr/Professor _____

Child's Name _____

Address _____

Contact Phone Numbers _____

- I have received a Parent/Guardian Information Statement to keep and I believe I understand the purpose, extent and possible effects of my child's involvement
- I have been asked if I would like to have a family member or friend with me while the project was explained
- I have had an opportunity to ask questions and I am satisfied with the answers I have received
- I understand that the researcher has agreed not to reveal results of any information involving my child, subject to legal requirements
- If information about this project is published or presented in any public form, I understand that the researcher will not reveal my child's identity
- I understand that if I refuse to consent to my child's participation, or if I withdraw my child from the project at any time without explanation, this will not affect my child's access to the best available treatment options and care from the Royal Children's Hospital and Health Services District.
- I understand I will receive a copy of this consent form

I give permission for the summary report of my child's progress from the study to be included in the hospital record (please tick): ❑ **yes** ❑ **no**

I am happy for my child to receive the Brain MRI scan under general anaesthesia after 24 months corrected age (please tick): ❑ **yes** ❑ **no**

SIGNATURE _____ **Date** _____

I have explained the study to the parent/guardian who has signed above, and believe that they understand the purpose, extent and possible effects of their child's involvement in this study.

RESEARCHER'S SIGNATURE _____ **Date** _____

Note: All parties signing the Consent Form must date their own signature.

Queensland cerebral palsy & rehabilitation research centre

Queensland Government
Queensland Health

Children's Health Service District - Royal Children's Hospital
PARENT/GUARDIAN INFORMATION STATEMENT
AND CONSENT FORM

Research Project Title: Queensland Cerebral Palsy Child Study: Nutrition, Growth and Physical Activity of Children

Researchers: Prof Peter Davies, Prof Roslyn Boyd, Dr Kristie Bell, Dr Sean Tweedy, Prof Richard Stevenson, Dr Stewart Trost, Dr Robert Ware, Ms Kelly Weir, Dr Lynne McKinlay, Dr Kate Sinclair, Christine Finn, Rachel Jordan, Jo-anne McMah, Jacqueline Walker, Katherine Benfer, Stina Oftedal and Laura Pareezer.

Thank you for taking the time to read this information Statement. This Information Statement and Consent Form is 6 pages long. Please make sure you read all pages.
For people who speak languages other English: If you would also like information about the research and a Consent Form in your language, please ask the person explaining this project to you.

You are invited to participate in the research project that is explained below.
What is an Information Statement?
These pages tell you about the research project. It explains to you all the steps and procedures of the project. The information is to help you decide whether or not you would like your child to take part in the research. Please read this Information Statement carefully. You are welcome to ask us questions about anything in it. You may wish to talk about the project with your family, friends or health care worker.

Participation in this research is entirely voluntary. If you don't want your child to take part, you don't have to. You can withdraw your child from the study at any time without explanation and there will be no penalty from any staff at the Royal Children's Hospital or the University of Queensland. Withdrawal will not affect your child's care in any way.

What is this research project about?
This project is about growth, nutrition, diet and physical activity of children who have cerebral palsy. Cerebral palsy is a physical disability caused by early brain injury. It occurs in 1 in 500 children. Children with cerebral palsy may be shorter and thinner than their typically developing peers. This project will look at how eating and drinking skills, dietary intake, and the amount of physical activity that children with cerebral palsy do effects the way they grow and develop, their quality of life, participation and the amount of health care used.

We are currently recruiting children with cerebral palsy aged 18-24 months to participate in a sub-study to improve the assessment tools used. Without an accurate way to measure children's eating and drinking, it is difficult to get a clear picture of how common it is and how it affects children and families. Mealtime videos of your child eating four foods (puree, semi-solid, soft chewable and biscuit) and a drink (from a cup and straw) will be analysed to see if there is any differences in scores between two mealtimes, between two raters, and between two ratings of the same video by a

single rater. The mealtime will be rated from video by Ms Katherine Benfer, PhD Student and Speech Pathologist, and Ms Kelly Weir, Senior Clinical Speech Pathologist, using three standardized measures; the Schedule for Oral Motor Assessment (SOMA), Dysphagia Disorders Survey (DDS), the Functional Feeding Assessment modified, as well as observing any signs of pharyngeal phase impairment (like coughing or gagging). The appointment will only take about 20-30 minutes, while your child is videoed having their snack, and can be completed at the Royal Children's Hospital or your home. Within a month of your first visit, a follow-up visit will be organised. This should be at the same place as your first assessment, and will take another 20-30 minutes.

Who are the researchers?

- Professor Peter Davies is the Director of the Children's Nutrition Research Centre at the University of Queensland.
- Professor Roslyn Boyd is a Paediatric Physiotherapist and Scientific Director at the Queensland Cerebral Palsy and Rehabilitation Research Centre, University of Queensland and the Royal Children's Hospital (Brisbane).
- Dr Kristie Bell is a Paediatric Dietician at the Royal Children's Hospital and the University of Queensland. She will coordinate the project and supervise the assessments.
- Professor Richard Stevenson is a Paediatrician at the Kluge Children's Rehabilitation Centre in the United States of America.
- Dr Sean Tweedy is an Exercise Physiologist at the University of Queensland.
- Ms Kelly Weir is a speech pathologist at the University of Queensland and the Royal Children's Hospital, she will analyse the video of your child's eating.
- Dr Lynne McKinlay is a Rehabilitation Specialist and Director of the Department of Rehabilitation at the Royal Childrens Hospital. She will discuss the the diagnosis of cerebral palsy.
- Dr Kate Sinclair is a neurologist at the Royal Children's Hosptial. She will discuss the diagnosis of cerebral palsy.
- Associate Professor Stewart Trost from the Department of Nutrition and Exercise Science, Oregon State University, will provide advice regarding the collection of the physical activity data.
- Dr Robert Ware is a statistician with the University of Queensland.

Why is my child being asked to be in this research project?

We are asking your child to take part because he/she has cerebral palsy and is aged between 18-24 months.

What are the alternatives to taking part in this project?

There is no obligation to participate in this project. Should you choose not to participate in this project, your child will have all the usual access to treatment at the Royal Children's Hospital and District Health Service.

What does my child need to do to be in this research project?

Your child needs to be present eating a one off snack, and they will be video-taped. This should last 20-30 minutes, and can be completed at home or another setting. During this visit they will need to eat:

1. Pureed food
2. Semi-solid food
3. Easy chew food (cheese-stick)
4. Biscuit (arrowroot biscuit)

Foods are to be provided by the family, apart from the cheese-stick and arrowroot biscuit. If your child is allergic to any of these foods, alternatives can be presented.

How will this study benefit my child?
You will be given a summary report of your child's performance based on their assessment results. The final study results will be summarized and reported back to you at the conclusion of the study.

How will this study benefit other people in the future?
The results of this study will provide valuable information that will help us to identify why some children with cerebral palsy grow poorly and how poor growth, dietary intake and physical activity may impact on their quality of life, participation and the amount of health care used. It will also assist us to determine which children need help to improve their nutrition, growth and physical activity and at what age is the best time to do this. In addition, it will provide us with information about how well different methods can measure the body composition of children with cerebral palsy and allow us to make recommendations on their use for others working with children with cerebral palsy.

What are the risks for my child?
There are no additional risks for your child with these measurements over and above that experienced in an everyday mealtime. The assessment is only observational, and safe.

What are the possible inconveniences?
The only inconvenience relates to you and your child's time, but assessments are relatively brief and can be scheduled at a time and location that suits you. We will pay for the cost of parking your car at the hospital, should you choose to attend here for the assessment.

What will be done to make sure the information is confidential?
Data from these assessments will be stored electronically without your child's name. A number will be used to identify them. This number will be linked to your child's name and the linking file will be kept confidential and only made available to the researchers. A separate database will contain your contact information and those results required for the generation of clinical reports. All databases will be password protected with limited access available to the researchers involved in the study.

Data collection sheets recording the assessments and the videotapes of the assessments will be stored in an individual file for your child in a secure, locked, fire proof filing cabinet. Only the researchers will have access to this information. These data sheets will be kept for 7 years at the Royal Children's Hospital. If we give talks or write about the results of this project, we will not use any names. All names and identifying information will be removed from data prior to any analysis.

Will I be informed of the results when the research project is finished?
If at any time you would like more information about your child's results, an appointment may be organized with one of the researchers. A regular newsletter will also be sent to you about the progress of the study. At the end of the study all families will be sent a summary of the results. The newsletters and final summary will talk about the children as a group and your child will not be identified in person.

You can decide whether or not to give permission for your child to take part in this research project. You can decide whether or not you would like to withdraw your child at any time

without explanation. Your decision whether or not for your child to participate will not prejudice your child's future relations with the Royal Children's Hospital and District Health Service. If you decide for your child to participate, you are free to withdraw your consent and discontinue participation at any time. The decision to withdraw from the study will not affect their routine medical treatment or their relationship with the people treating them. You may like to discuss your child's participation in this research project with your family and with your doctor. You can ask for further information before deciding to take part.

If you would like more information about the study or if you need to contact a study representative in an emergency, the person to contact is:

Name: Prof Roslyn Boyd, Scientific Director Cerebral Palsy and Rehabilitation Research,
Contact telephone: (07) 3646 5315 **mobile:-** 0434 608 443
Or
Name: Ms Katherine Benfer, PhD Scholar and Speech Pathologist
Contact telephone: (07) 3646 5442.

What are my child's rights as a participant?
I am informed that except where stated above, no information regarding my child's medical history will be released. This is subject to legal requirements. I am informed that the results of any tests involving my child will not be published so as to reveal my child's identity. This is subject to legal requirements. The detail of the procedure proposed has also been explained to me. This includes how long it will take, how often the procedure will be performed and whether any discomfort will result. It has also been explained that my child's involvement in the research may not be of any benefit to him or her. I understand that the purpose of this research project is to improve the quality of medical care in the future. I have been asked if I would like to have a family member or a friend with me while the project is explained to me. I understand that this project follows the guidelines of the National Statement on Ethical Conduct in Research Involving Humans (1999).I understand that this research project has been approved by the Royal Children's Hospital Ethics Committee on behalf of the Royal Children's Hospital and Health Services District, Brisbane. I have received a copy of this document.

Contact:-
The Research Ethics Committee of the Royal Children's Hospital and Health Services District has approved this study. Should you wish to discuss the study with someone not directly involved, in particular in relation to matters concerning policies, information about the conduct of the study or your rights as a participant, or you wish to make a confidential complaint, at any time, you may contact the Co-ordinator on the ethics committee, Royal Childrens Hospital and Health Services District, c/o Dept of Pediatrics and Child Health, Level 3, Foundation building, Herston. QLD. 4029. This study adheres to the Guidelines of the ethical review process of The University of Queensland. Whilst you are free to discuss your participation in this study with project staff (contactable on 07 3636 5542), if you would like to speak to an officer of the University not involved in the study, you may contact the Ethics Officer on 3365 3924.

This study has been approved by the Cerebral Palsy League of Queensland Ethics Committee (**CPLQ- 2009/10 – 1029**). If needed, verification can be obtained by writing or telephoning the Cerebral Palsy League Ethics Committee, c/-Cerebral Palsy League of Queensland, 55 Oxlade Drive, New Farm, Brisbane Qld 4005 or PO Box 386, Fortitude Valley QLD 4006 Tel: 07 33588101

STANDARD INFORMED CONSENT FOR PARENT/GUARDIAN TO GIVE CONSENT FOR THEIR CHILD TO PARTICIPATE IN A RESEARCH PROJECT

Project Number

EHRC No 2008002260

Title of Project

QLD CP Child study: Nutrition, Growth and Physical Activity

Investigator(s)

Prof Peter Davies, Prof Roslyn Boyd, Dr Kristie Bell, Dr Sean Tweedy, Prof Richard Stevenson, Dr Stewart Trost, Dr Robert Ware, Ms Kelly Weir, Dr Lynne McKinlay, Dr Kate Sinclair, Christine Finn, Rachel Jordan, Lauren Forbes, Jacqueline Walker, Katherine Benfer, Stina Oftedal and Laura Pareezer.

I (Parent/Guardian name) _____

voluntarily consent for my child to take part in the above titled Research Project, explained to me by

Mr/Ms/Dr/Professor _____

Child's Name _____

Address _____

Contact Phone Numbers _____

- I have received a Parent/Guardian Information Statement to keep and I believe I understand the purpose, extent and possible effects of my child's involvement
- I have been asked if I would like to have a family member or friend with me while the project was explained
- I have had an opportunity to ask questions and I am satisfied with the answers I have received
- I understand that the researcher has agreed not to reveal results of any information involving my child, subject to legal requirements
- If information about this project is published or presented in any public form, I understand that the researcher will not reveal my child's identity
- I understand that if I refuse to consent to my child's participation, or if I withdraw my child from the project at any time without explanation, this will not affect my child's access to the best available treatment options and care from the Royal Children's Hospital and Health Services District.
- I understand I will receive a copy of this consent form

I give permission for the summary report of my child's progress from the study to be included in the hospital record (please tick): ❑ yes ❑ no

SIGNATURE _____ **Date** _____

I have explained the study to the parent/guardian who has signed above, and believe that they understand the purpose, extent and possible effects of their child's involvement in this study.

RESEARCHER'S SIGNATURE _____ **Date** _____

Note: All parties signing the Consent Form must date their own signature.

Queensland cerebral palsy & rehabilitation research centre

Queensland Government
Queensland Health

Children's Health Service District - Royal Children's Hospital
PARENT/GUARDIAN INFORMATION STATEMENT
AND CONSENT FORM

Research Project Title: Queensland Cerebral Palsy Child Study: Nutrition, Growth and Physical Activity of Children

Researchers: Prof Peter Davies, Prof Roslyn Boyd, Dr Kristie Bell, Dr Sean Tweedy, Prof Richard Stevenson, Dr Stewart Trost, Dr Robert Ware, Ms Kelly Weir, Dr Lynne McKinlay, Dr Kate Sinclair, Christine Finn, Rachel Jordan, Jo-anne McMah, Jacqueline Walker, Katherine Benfer, Stina Oftedal and Laura Pareezer.

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You are invited to participate in the research project that is explained below.
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Participation in this research is entirely voluntary. If you don't want your child to take part, you don't have to. You can withdraw your child from the study at any time without explanation and there will be no penalty from any staff at the Royal Children's Hospital or the University of Queensland. Withdrawal will not affect your child's care in any way.

What is this research project about?
This project is about growth, nutrition, diet and physical activity of children who have cerebral palsy. Cerebral palsy is a physical disability caused by early brain injury. It occurs in 1 in 500 children. Children with cerebral palsy may be shorter and thinner than their typically developing peers. This project will look at how eating and drinking skills, dietary intake, and the amount of physical activity that children with cerebral palsy do effects the way they grow and develop, their quality of life, participation and the amount of health care used.

We are currently recruiting a reference group of children aged 18-36 months, born full term (>37 weeks), with no admissions to the neonatal care unit, no diagnosis receiving medical/ allied health care, and not on regular medications. Participation involves two sub-studies:
<u>Oral motor and swallowing skills:</u>
Understanding the oral motor and swallowing skills of children with typical feeding skills and their performance on three specific assessments will help strengthen these measures, which are being

used in the cerebral palsy study. The mealtime will be rated from video by Ms Katherine Benfer, PhD Student and Speech Pathologist, using three standardized measures; the Schedule for Oral Motor Assessment (SOMA), Dysphagia Disorders Survey (DDS) and the Functional Feeding Assessment modified (FFAm) as well as observing any signs of pharyngeal phase impairment (like coughing or gagging). Children will be taped eating four food textures (puree, semi-solid, soft chewable and biscuit) and drinking (from their typical cup and a straw). The appointment will only take about 20-30 minutes, and can be completed at your home or the Royal Children's Hospital.

Levels of physical activity:

Being able to measure the amount and intensity of physical activity undertaken by toddlers who are typically developing will helps us understand whether children with cerebral palsy differ from their typically developing peers in terms of the amount of physical activity they participate in. Day-to-day physical activity will be measured by a small, lightweight (27g) activity monitor, which your child will wear around their waist during waking hours for three days. To calibrate the activity monitor for use this age group, your child will also be videotaped while wearing it during their mealtime assessment and for 10-15 minutes while playing. Videos will then be rated by Stina Oftedal, PhD student and Dietitian. The assessment will only add 15 minutes to the mealtime assessment. You will also be given an activity monitor to take home, which a courier will pick up upon completion of the three- day wear period.

Who are the researchers?

- Professor Peter Davies is the Director of the Children's Nutrition Research Centre at the University of Queensland.
- Professor Roslyn Boyd is a Paediatric Physiotherapist and Scientific Director at the Queensland Cerebral Palsy and Rehabilitation Research Centre, University of Queensland and the Royal Children's Hospital (Brisbane).
- Dr Kristie Bell is a Paediatric Dietician at the Royal Children's Hospital and the University of Queensland. She will coordinate the project and supervise the assessments.
- Professor Richard Stevenson is a Paediatrician at the Kluge Children's Rehabilitation Centre in the United States of America.
- Dr Sean Tweedy is an Exercise Physiologist at the University of Queensland.
- Ms Kelly Weir is a speech pathologist at the University of Queensland and the Royal Children's Hospital, she will analyse the video of your child's eating.
- Dr Lynne McKinlay is a Rehabilitation Specialist and Director of the Department of Rehabilitation at the Royal Childrens Hospital. She will discuss the the diagnosis of cerebral palsy.
- Dr Kate Sinclair is a neurologist at the Royal Children's Hosptial. She will discuss the diagnosis of cerebral palsy.
- Associate Professor Stewart Trost from the Department of Nutrition and Exercise Science, Oregon State University, will provide advice regarding the collection of the physical activity data.
- Dr Robert Ware is a statistician with the University of Queensland.

Why is my child being asked to be in this research project?
We are asking your child to take part because he/she is 18-36 months, born full term (<37 weeks), with no admissions to the neonatal care unit, no diagnosis receiving medical/ allied health care, and not on regular medications.

What are the alternatives to taking part in this project?
There is no obligation to participate in this project. Should you choose not to participate in this project, your child will have all the usual access to treatment at the Royal Children's Hospital and District Health Service.

What does my child need to do to be in this research project?
Mealtime assessment
Your child needs to be present eating a one off snack, and they will be video-taped. This should last 20-30 minutes, and can be completed at home or another setting. During this visit they will need to eat:

1. Pureed food
2. Semi-solid food
3. Easy chew food (cheese-stick)
4. Biscuit (arrowroot biscuit)

Foods are to be provided by the family, apart from the cheese-stick and arrowroot biscuit. If your child is allergic to any of these foods, alternatives can be presented.

Physical activity level
To calibrate the activity monitor your child needs to wear it during the mealtime assessment and for 10-15 minutes of playtime where they will be videotaped. The activity monitor is attached around their waist on a soft belt. Your child will also need to wear the activity monitor for three days. This does not need to be directly after the appointment, and the days do not have to be consecutive. The activity monitor can be placed under clothes and is taken off during sleep and for water activities. An activity log is filled out by a parent to indicate when the monitor is put on and taken off during the three days.

Anthropometry
The height, weight and body composition (fat mass and lean mass) of your child will also be measured. Body composition is measured using bioelectrical impedance analysis which is a simple, painless and safe technique. The technique requires that your child lie quietly with surface electrodes taped lightly to their wrist and ankle. A very small electrical current passes through the body, which is completely safe and so mild it cannot be felt. The measurement only takes a few seconds during which your child will not be able to wear shoes, socks and metallic jewelry.

How will this study benefit my child?
The study will not have any direct benefits to you or your child.

How will this study benefit other people in the future?
The results of this study will provide valuable information that will help us to identify why some children with cerebral palsy grow poorly and how poor growth, dietary intake and physical activity may impact on their quality of life, participation and the amount of health care used. It will also assist us to determine which children need help to improve their nutrition, growth and physical activity and at what age is the best time to do this. In addition, it will provide us with information

about how well different methods can measure the body composition of children with cerebral palsy and allow us to make recommendations on their use for others working with children with cerebral palsy.

What are the risks for my child?
There are no additional risks for your child with these measurements over and above that experienced in an everyday mealtime and play. The assessment is only observational, and safe.

What are the possible inconveniences?
The only inconvenience relates to you and your child's time, but assessments are relatively brief and can be scheduled at a time and location that suits you. We will pay for the cost of parking your car at the hospital, should you choose to attend the Royal Children's Hospital for the assessment.

What will be done to make sure the information is confidential?
Data from these assessments will be stored electronically without your child's name. A number will be used to identify them. This number will be linked to your child's name and the linking file will be kept confidential and only made available to the researchers. A separate database will contain your contact information and those results required for the generation of clinical reports. All databases will be password protected with limited access available to the researchers involved in the study.

Data collection sheets recording the assessments and the videotapes of the assessments will be stored in an individual file for your child in a secure, locked, fire proof filing cabinet. Only the researchers will have access to this information. These data sheets will be kept for 7 years at the Royal Children's Hospital. If we give talks or write about the results of this project, we will not use any names.

All names and identifying information will be removed from data prior to any analysis.

Will I be informed of the results when the research project is finished?
A regular newsletter will also be sent to you about the progress of the study. At the end of the study all families will be sent a summary of the results. The newsletters and final summary will talk about the children as a group and your child will not be identified in person.

You can decide whether or not to give permission for your child to take part in this research project. You can decide whether or not you would like to withdraw your child at any time without explanation. Your decision whether or not for your child to participate will not prejudice your child's future relations with the Royal Children's Hospital and District Health Service. If you decide for your child to participate, you are free to withdraw your consent and discontinue participation at any time. The decision to withdraw from the study will not affect their routine medical treatment or their relationship with the people treating them. You may like to discuss your child's participation in this research project with your family and with your doctor. You can ask for further information before deciding to take part.

If you would like more information about the study or if you need to contact a study representative in an emergency, the person to contact is:

Name: Prof Roslyn Boyd, Scientific Director Cerebral Palsy and Rehabilitation Research,
Contact telephone: (07) 3365 5315 **mobile:-** 0434 608 443

Or

Name: Dr Kristie Bell, Clinical Postdoctoral Research Fellow
Contact telephone: (07) 3646 5537.

What are my child's rights as a participant?
I am informed that except where stated above, no information regarding my child's medical history will be released. This is subject to legal requirements. I am informed that the results of any tests involving my child will not be published so as to reveal my child's identity. This is subject to legal requirements. The detail of the procedure proposed has also been explained to me. This includes how long it will take, how often the procedure will be performed and whether any discomfort will result. It has also been explained that my child's involvement in the research may not be of any benefit to him or her. I understand that the purpose of this research project is to improve the quality of medical care in the future. I have been asked if I would like to have a family member or a friend with me while the project is explained to me. I understand that this project follows the guidelines of the National Statement on Ethical Conduct in Research Involving Humans (1999).I understand that this research project has been approved by the Royal Children's Hospital Ethics Committee on behalf of the Royal Children's Hospital and Health Services District, Brisbane. I have received a copy of this document.

Contact:-
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This study has been approved by the Cerebral Palsy League of Queensland Ethics Committee (**CPLQ- 2009/10 – 1029**). If needed, verification can be obtained by writing or telephoning the Cerebral Palsy League Ethics Committee, c/-Cerebral Palsy League of Queensland, 55 Oxlade Drive, New Farm, Brisbane Qld 4005 or PO Box 386, Fortitude Valley QLD 4006 Tel: 07 33588101

STANDARD INFORMED CONSENT FOR PARENT/GUARDIAN TO GIVE CONSENT FOR THEIR CHILD TO PARTICIPATE IN A RESEARCH PROJECT

Project Number

EHRC No 2008002260

Title of Project

QLD CP Child study: Nutrition, Growth and Physical Activity

Investigator(s)

Prof Peter Davies, Prof Roslyn Boyd, Dr Kristie Bell, Dr Sean Tweedy, Prof Richard Stevenson, Dr Stewart Trost, Dr Robert Ware, Ms Kelly Weir, Dr Lynne McKinlay, Dr Kate Sinclair, Christine Finn, Rachel Jordan, Jo-anne McMah, Jacqueline Walker, Katherine Benfer, Stina Oftedal and Laura Pareezer.

I (Parent/Guardian name) _____

voluntarily consent for my child to take part in the above titled Research Project, explained to me by

Mr/Ms/Dr/Professor _____

Child's Name _____

Address _____

Contact Phone Numbers _____

- I have received a Parent/Guardian Information Statement to keep and I believe I understand the purpose, extent and possible effects of my child's involvement
- I have been asked if I would like to have a family member or friend with me while the project was explained
- I have had an opportunity to ask questions and I am satisfied with the answers I have received
- I understand that the researcher has agreed not to reveal results of any information involving my child, subject to legal requirements
- If information about this project is published or presented in any public form, I understand that the researcher will not reveal my child's identity
- I understand that if I refuse to consent to my child's participation, or if I withdraw my child from the project at any time without explanation, this will not affect my child's access to the best available treatment options and care from the Royal Children's Hospital and Health Services District.
- I understand I will receive a copy of this consent form

I give permission for the summary report of my child's progress from the study to be included in the hospital record (please tick): ❑ yes ❑ no

SIGNATURE _____ **Date** _____

I have explained the study to the parent/guardian who has signed above, and believe that they understand the purpose, extent and possible effects of their child's involvement in this study.

RESEARCHER'S SIGNATURE _____ **Date** _____

Note: All parties signing the Consent Form must date their own signature.

Queensland cerebral palsy & rehabilitation research centre

Children's Health Service District - Royal Children's Hospital
PARENT/GUARDIAN INFORMATION STATEMENT
AND CONSENT FORM

Research Project Title: Queensland Cerebral Palsy Child Study: Nutrition, Growth and Physical Activity of Children – Oropharyngeal Dysphagia in Bangladesh (OPD-Bd)

Researchers: Ms Katherine Benfer, Prof Roslyn Boyd, Ms Kelly Weir, Prof Peter Davies, Dr Kristie Bell, Dr Robert Ware, Md Jahangir Alam, Dr Baitun Nahar, Dr Sabera Bilkis, Ms Hosneara Perveen, Ms Fatema Akhter Mitu, Dr Sasaka Bandaranyake, Ms Laura Pareezer, Ms Rachel Jordan, Ms Christine Finn, Prof Thameed Ahmed.

You are invited to participate in the research project that is explained below.
What is an Information Statement?
These pages tell you about the research project. It explains to you all the steps and procedures of the project. The information is to help you decide whether or not you would like your child to take part in the research. Please read this Information Statement carefully. You are welcome to ask us questions about anything in it. You may wish to talk about the project with your family, friends or health care worker.

Participation in this research is entirely voluntary. If you don't want your child to take part, you don't have to. You can withdraw your child from the study at any time without explanation and there will be no penalty from any staff at the Centre for Rehabilitation of the Paralysed. Withdrawal will not affect your child's care in any way.

What is this research project about?
This project is about feeding, diet and growth of children who have cerebral palsy. Cerebral palsy is a physical disability caused by early brain injury. Children with cerebral palsy may be shorter and thinner than their typically developing peers. This project will look at how eating and drinking skills, dietary intake, and other factors affects the way they grow and develop.

Who are the researchers?
- Ms Katherine Benfer is a PhD student and speech pathologist with the Queensland Cerebral Palsy and Rehabilitation Research Centre, The University of Queensland.
- Professor Roslyn Boyd is a Paediatric Physiotherapist and Scientific Director at the Queensland Cerebral Palsy and Rehabilitation Research Centre, University of Queensland and the Royal Children's Hospital (Brisbane).
- Ms Kelly Weir is a speech pathologist at the University of Queensland and the Royal Children's Hospital, she will analyse the video of your child's eating.
- Professor Peter Davies is the Director of the Children's Nutrition Research Centre at the University of Queensland.
- Dr Kristie Bell is a Paediatric Dietician at the Royal Children's Hospital and the University of Queensland. She will coordinate the project and supervise the assessments.
- Dr Robert Ware is a statistician with the University of Queensland.
- Md. Jahangir Alam is the Course Coordinator at the Bangladesh Health Professions Institute, Centre for Rehabilitation of the Paralysed. He is the main Bangladeshi contact for the study. In addition, Ms

Hosneara Perveen and Ms Fatema Akhter Mitu (Centre for Rehabilitation of the Paralysed) will assist in the research in Bangladesh.

- Dr Sabera Bilkis (Centre for Rehabilitation of the Paralysed) and Dr Sasaka Bandaranyake (Royal Children's Hospital, Brisbane Australia) will provide the diagnosis of cerebral palsy.
- Ms Rachel Jordan and Ms Christine Finn are physiotherapists from the Queensland Cerebral Palsy and Rehabilitation Research Centre. They will rate your child's severity and type of cerebral palsy.
- Dr Baitun Nahar and Dr Tahmeed Ahmed are researchers from ICDDR,B (International Centre for Diarrhoeal Disease Research Centre). They will be involved in conducting research in the Bangladeshi context.

Why is my child being asked to be in this research project?
We are asking your child to take part because he/she has cerebral palsy and is aged between 18-36 months.

What are the alternatives to taking part in this project?
There is no obligation to participate in this project. Should you choose not to participate in this project, your child will have all the usual access to treatment at the Centre for Rehabilitation of the Paralysed.

What does my child need to do to be in this research project?
There may be 2 interviews with you (for about an hour each), a one hour session with your child, and one day when the researcher will help with weighing your child's food.

Answering some questions with your doctor, the researcher and a translator, about:

- Your household and community
- Your child's birth, and development
- How and what your child eats
- Medical services you access

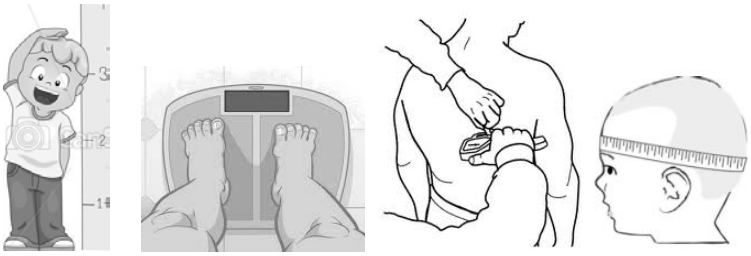
Video-taping your child eating and drinking

3 spoonfuls of these foods:





Measuring your child's growth



Measuring your height and weight

Measuring your child's body composition

To do this your child will need to lie still while we stick electrodes on their hands and feet, which are connected to a bioelectrical impedance machine. A small current goes through their body, but they don't feel anything.

Video-taping your child moving and playing

Weighing what your child eats in a day

How will this study benefit my child?
The results of the comprehensive assessment will be given to your speech therapist, which will give them information to assist in planning treatment goals and techniques.

How will this study benefit other people in the future?
The results of this study will provide valuable information that will help us to identify why some children with cerebral palsy have trouble with eating and grow poorly. It will also assist us to determine which children need help to improve their nutrition, at what age is the best time to do this and what strategies may help the most

What are the risks for my child?
There are no additional risks for your child with these measurements over and above that experienced in an everyday mealtime. The mealtime assessment is only observational, and safe. The growth measurements may be uncomfortable for some children, but are safe and pain free.

What are the possible inconveniences?
The only inconvenience relates to you and your child's time, but assessments can be scheduled around your therapy and other appointments at CRP at a time that suits you.

What will be done to make sure the information is confidential?
Data from these assessments will be stored electronically without your child's name. All databases will be password protected with limited access available to the researchers involved in the study.

Data collection sheets recording the assessments and the videotapes of the assessments will be de-identified and stored electronically. This may be stored short term on an external harddrive which will be stored in a secure, locked filing cabinet in the Paediatric Unit. Longer term, files will be transferred over to The University of Queensland share drive. Only the researchers will have access to this information. These data sheets will be kept for 7 years at the Royal Children's Hospital. If we give talks or write about the results of this project, we will not use any names. All names and identifying information will be removed from data prior to any analysis.

Will I be informed of the results when the research project is finished?
If at any time you would like more information about your child's results, an appointment may be organised with one of the researchers.

- **You can decide whether or not to give permission for your child to take part in this research project.**
- **You can decide whether or not you would like to withdraw your child at any time without explanation.**
- **Your decision to not participate or withdraw from the study will not affect your routine medical treatment, your relationship with the people treating them or any of the services you receive from the Centre for Rehabilitation of the Paralysed.**
- **You may like to discuss your child's participation in this research project with your family and with your doctor.**
- **You can ask for further information before deciding to take part.**

For more information about the study, please contact:
Name: Ms Katherine Benfer, PhD Scholar and Speech Pathologist
Contact telephone: TBC
Or
Name: Md Jahangir Alam, Centre for Rehabilitation of the Paralysed
Contact telephone: 01716637992

Complaints or ethical issues:
Should you wish to discuss the study with someone not directly involved, in particular in relation to matters concerning policies, information about the conduct of the study or your rights as a participant, or you wish to make a confidential complaint, at any time, you may contact an ethics officer of one of the ethics committees responsible:

- Royal Children's Hospital and Health Services District, c/o Dept of Pediatrics and Child Health, Level 3, Foundation building, Herston. QLD. 4029.
- Ms Reshma Parvin, Research Associate/ Ethics Officer, Centre for Rehabilitation of the Paralysed: +88-02-7745464-5
- Salam Khan, IRB Secretariat, Research Review Committee, ICDDR,B: +88 02 9886498

This study adheres to the Guidelines of the ethical review process of The University of Queensland. The Research Ethics Committee of the Royal Children's Hospital and Health Services District, Human Ethics Research Committee of the Centre for Rehabilitation of the Paralysed and ICDDR,B have approved this study.

Queensland cerebral palsy & rehabilitation research centre

STANDARD INFORMED CONSENT FOR PARENT/GUARDIAN TO GIVE CONSENT FOR THEIR CHILD TO PARTICIPATE IN A RESEARCH PROJECT

Project Number

EHRC No 2008002260

Title of Project

QLD CP Child study: Nutrition, Growth and Physical Activity – Oropharyngeal Dysphagia in Bangladesh

Investigator(s)

Ms Katherine Benfer, Prof Roslyn Boyd, Ms Kelly Weir, Prof Peter Davies, Dr Kristie Bell, Dr Robert Ware, Md Jahangir Alam, Dr Baitun Nahar, Dr Sabera Bilkis, Ms Hosneara Perveen, Ms Fatema Akhter Mitu, Dr Sasaka Bandaranyake, Ms Laura Pareezer, Ms Rachel Jordan, Ms Christine Finn, Prof Thameed Ahmed

I (Parent/Guardian name) _____

voluntarily consent for my child to take part in the above titled Research Project, explained to me by

Mr/Ms/Dr/Professor _____

Child's Name _____

Address _____

Contact Phone Numbers _____

- I have received a Parent/Guardian Information Statement to keep and I believe I understand the purpose, extent and possible effects of my child's involvement
- I have been asked if I would like to have a family member or friend with me while the project was explained
- I have had an opportunity to ask questions and I am satisfied with the answers I have received
- I understand that the researcher has agreed not to reveal results of any information involving my child, subject to legal requirements
- If information about this project is published or presented in any public form, I understand that the researcher will not reveal my child's identity
- I understand that if I refuse to consent to my child's participation, or if I withdraw my child from the project at any time without explanation, this will not affect my child's access to the best available treatment options and care from the Centre for Rehabilitation of the Paralysed.
- I understand I will receive a copy of this consent form

I give permission for the results of my child's assessment to be included in their Paediatric Unit file and discussed with their primary speech therapist: (please tick): ❑ yes ❑ no

SIGNATURE _____ **Date** _____

I have explained the study to the parent/guardian who has signed above, and believe that they understand the purpose, extent and possible effects of their child's involvement in this study.

RESEARCHER'S SIGNATURE _____ **Date** _____

Note: All parties signing the Consent Form must date their own signature.

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The University of Queensland

Qld CPchild: Brain Function & Motor Development Study
The class of 2006-2009

Can you help us?
Researchers from the Queensland Cerebral Palsy & Rehabilitation Research Centre (at the Royal Children's Hospital) are looking for **all children** with **Cerebral Palsy born in Queensland** in the **birth years of 2006, 2007, 2009 and 2009.** Please note your child can enter the study at any age.

The study will measure your child's motor development, muscle and bone development and see if these are related to the nature of the brain injury they have sustained. Information from the study will help many children with Cerebral Palsy and their families in the future. The information will allow us to learn more about the specific needs and potential outcomes of children with Cerebral Palsy.

Benefits: Your child will receive regular, comprehensive surveillance. All information will be reported back to you after each visit and the results sent to your child's paediatrician and therapists to keep them informed of your child's progress. If you child has not had a brain MRI we would discuss with you how this would be helpful.

The study involves 6 visits over 4 years to the Royal Children's Hospital or your regional hospital (whichever is more convenient). These visits will be performed when your child is 18, 24, 30 and 36 months of age, and then around their 4th and 5th birthdays. Each visit takes about 1 ½ - 2 hours.

Qld CPchild: Growth, Nutrition & Physical Activity Study The class of 2006-2009

Another study is being conducted in conjunction with the Qld CP Child study, and researchers are looking for **all children** with **cerebral palsy born in Queensland** in the **birth years of 2006 - 2009.** Your child can enter the study at any time from 18 months to 5 years.

The study will measure your child's growth, nutrition, diet and physical activity and see if these relate to health outcomes, participation and health related quality of life.

Benefits: Your child will receive regular, comprehensive surveillance of their growth, nutrition and physical activity, with all information reported back to you and your therapists as mentioned above.

The study involves 3 visits over 4 years to the Royal Children's Hospital or your regional hospital. These visits will be performed when your child is 18-24 months, and then around their 3rd and 5th birthdays. These visits will coincide with assessments for the Qld CP Child study. Each visit takes about 2 - 2.5 hours in total.

If you would like to find out more about either study please contact either:

Rachel Jordan Study Coordinator and Physiotherapist, (07) 3636 5665, Rachel_Jordan1@health.qld.gov.au

Dr Kristie Bell, Paediatric Dietitian & Growth, Nutrition & Physical Activity Study Coordinator,
(07) 3636 5542, kristie_bell@health.qld.gov.au

Laura Pareezer, Clinical Nurse Consultant, Clinical Trials, (07) 3636 5061, laura_pareezer@health.qld.gov.au

Professor Roslyn Boyd, Scientific Director, QCPRRC, 0434608443, r.boyd@uq.edu.au

Queensland cerebral palsy & rehabilitation research centre

QUEENSLAND CEREBRAL PALSY CHILD STUDIES

Qld CP Child: Growth, Nutrition & Physical Activity Feeding Study: Assessment Test Development Children with Cerebral Palsy

Can you help us?
Researchers from the Queensland Cerebral Palsy & Rehabilitation Research Centre (at the Royal Children's Hospital) are looking for **children aged 18-24 months** with **Cerebral Palsy.**

The study will measure your child's oral motor skills in feeding to determine the accuracy of the assessment tools in detecting feeding difficulties. You will get a summary report of your child's performance based on these assessments.

Information from the study will help many children with Cerebral Palsy and their families in the future. The information will allow us to learn more about the specific needs and potential outcomes of children with Cerebral Palsy.

The study involves 2 appointments of 20-30 minutes, either at the Royal Children's Hospital or your home. During the appointment your child will be videoed eating four foods (a puree like yoghurt; a lumpy/mashed food like mashed veges or baked beans; a cheesestick and a biscuit) and having a drink (from a cup and straw). The cheesestick and biscuit will be provided, but an alternative can be given if your child is allergic to either of these foods.

If you would like to find out more about this study please return the enclosed expression of interest form or contact:

Kath Benfer, Speech Pathologist and PhD student
Phone: (07) 3646 5542, Email: katherine.benfer@uqconnect.edu.au

Laura Pareezer, Clinical Nurse Consultant, Clinical Trials
Phone: (07) 3646 5061, Email: laura_pareezer@health.qld.gov.au

QLD Cerebral Palsy & Rehabilitation Research Centre, Royal Children's Hospital, Herston
Ph: (07) 3646 5542 Fax: (07) 3646 5538 Email: qcprrc@uq.edu.au

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RECRUITING TYPICALLY DEVELOPING PRESCHOOL CHILDREN

Qld CP Child: Growth, Nutrition & Physical Activity

Feeding & Physical Activity Studies

Can you help us?
Researchers from the Queensland Cerebral Palsy & Rehabilitation Research Centre (at the Royal Children's Hospital) are looking for **children aged 2.5-3 years** born full term (>37 weeks), with no admissions to the neonatal care unit, no diagnosis receiving medical/ allied health care, and not on regular medications**.**

The study will measure your child's oral motor skills in feeding to determine the accuracy of the assessment tools in detecting feeding difficulties in children with cerebral palsy. It will also measure your child's day to day physical activity level to allow us to assess if children with cerebral palsy differ in how much physical activity they participate in.

Information from the study will help many children with Cerebral Palsy and their families in the future. The information will allow us to learn more about the specific needs and potential outcomes of children with Cerebral Palsy.

The study involves a single appointment of 40-60 minutes either at the Royal Children's Hospital or your home. It will consist of two parts plus a home activity:

1. Mealtime assessment (20-30 minutes): During the appointment your child will be videoed eating four foods (a puree like yoghurt; a lumpy/mashed food like mashed veges or baked beans; a cheesestick and a biscuit) and having a drink (from a cup and straw). The cheesestick and biscuit will be provided, but an alternative can be given if your child is allergic to either of these foods.
2. Activity assessment (20-30 minutes): During the appointment your child will wear a small activity monitor around their waist while eating and while playing for 10-15 minutes while videotaped. Your child's height and weight will also be measured.
3. Home activity monitoring: Your child will wear the activity monitor for three days during waking hours (except water activities). A courier will pick up the monitor on completion.

If you would like to find out more about this study please contact:

Kath Benfer, Speech Pathologist and PhD student
Phone: (07) 3646 5372, Email: katherine.benfer@uqconnect.edu.au

Stina Oftedal, Dietitian and PhD student
Phone : (07) 3646 5372, Email : s.oftedal@uq.edu.au

Laura Pareezer, Clinical Nurse Consultant, Clinical Trials
Phone: (07) 3646 5061, Email: laura_pareezer@health.qld.gov.au

QLD Cerebral Palsy & Rehabilitation Research Centre, Royal Children's Hospital, Herston
Ph: (07) 3646 5542 Fax: (07) 3646 5538 Email: qcprrc@uq.edu.au

Queensland cerebral palsy & rehabilitation research centre

RECRUITING CHILDREN WITH CEREBRAL PALSY AGED 18-36 MONTHS

Qld CP Child: Growth, Nutrition & Physical Activity
Oropharyngeal Dysphagia in Bangladesh

Can you help us?
Researchers from the Queensland Cerebral Palsy & Rehabilitation Research Centre (at the Royal Children's Hospital) are looking for **children aged 18-36 months** with Cerebral Palsy

The study will measure your child's oral motor skills in feeding, dietary intake and nutritional status to determine ways to better help children with cerebral palsy grow.

Information from the study will help your speech therapist with the therapy program they give you. It will also help many children with Cerebral Palsy and their families in the future.

The study involves

Answering some questions about where you live, your child's birth, and how they eat

Videotaping your child eating

Queensland cerebral palsy & rehabilitation research centre

Measuring your child's growth
Having your own height and weight measured

Weighing what your child eats in a day

If you would like to find out more about this study please contact:

Kath Benfer, Speech Pathologist and PhD student
Phone: +617 3646 5372

Ms Hosneara Provan, Head of Paediatric Unit
Phone: +88 0277454645

Md Jahingur Alam, Course Coordinator Speech & Language Therapy
Ph: +88 0277454645 or 01716637992

QLD CP Child: Growth, Nutrition and Physical Activity: Snack Evaluation Protocol

Prior to Evaluation:
Parents are sent a parent explanation letter and the Children's Feeding Skills Questionnaire. The explanation letter will detail information about what types of food to bring for the 'snack video' and how we will video the child.

Foods to bring along:

- Smooth spoonable food: yoghurt, fruche, snack pac, puree fruit.
- Lumpy mashed spoonable food: e.g. yoghurt with muesli, lumpy vegetables, spaghetti oops, baked beans.
- Chewable food: biscuit, sandwich, fruit or muesli bar.
- Drink: Any type of drink.
- Utensils: the child's regular utensils (e.g. spoon, cups, spout/straw cups, pop tops, etc)

The mother/primary carer will feed the child if appropriate.

Snack Evaluation:
Videoing:

1. Camera set up & begin videotape:
    - The camera is set up to include a view of the child's face and neck. The view should include the whole of the child's head and a little of the upper chest/neck. If the mother/carer is feeding the child, angle the camera to the side of the shoulder of the hand that is not feeding the child (that way the view of the child's mouth is not obscured by the presentation of the food). The camera person may have to adjust the camera slightly to get a view of the child's mouth when drinking.
    - Ensure the child is well positioned.
    - Start the camera recording.
    - Prior to each texture a longer range view should be quickly taken to observe the child's position (either in chair, stroller, carer's arms etc) and keep it there for the first bite/mouthful. This will allow us to record the child's position, type of utensil and feeding independence.
    - Then do the close up position (head and neck) for the actual eating of most of the food (from the second bite to the finish).
    - NOTE: When the child is drinking from a bottle or cup: you will need to change the camera angle to be able to see the lips side on or from an above shot.

2. Pre-feeding check:
    - Complete the first page of the data sheet:
        - Mother/carer needs to write down what foods they have for each texture/consistency.
        - Sign the section about having prepared the food for their child, giving the food to their child and allergy information.

    - Check the following items and mark on the 'Snack Evaluation Sheet'

- **'Wet/gurgly breathing'** item:
  The researcher places his/her hands lightly on front and back of the child's chest. Ask the child take a big breath. (Model taking a big breath, or get parent to model taking a big breath – especially for younger age group.) Feel for 'gurgly breathing' / vibration / 'rattly chest' / fremitus. Mark the 'wet/gurgly breathing' item on snack evaluation sheet.

- '**Wet/gurgly voice'** item:
  Ask the child to say 'ahhhhh' in a big loud voice. If the child refuses to say ahh, ask the child/get carer to ask the child to do some counting. Listen to the voice quality and determine whether it is clear or wet/gurgly sounding. Mark the 'wet/gurgly voice' item on the snack evaluation sheet.

- **'Cough'** item:
  Observed the child and determine whether a spontaneous cough has been heard during the session. Mark the 'cough' item on the snack evaluation sheet.

- **'Drooling'** item.
  Observe the child and his/her garments. Mark the drooling severity score on the snack evaluation sheet.

Severity Scale
1. Dry – never drools
2. Mild – only lips wet
3. Moderate – wet on lips and chin
4. Severe – drools to the extent that clothes and/or objects get wet
5. Profuse – clothing, hands and objects become very wet

3. Instructions to the feeder:
   - Instruct the primary carer to present at least 3 presentations/bites of each type of food and drink [preferred order includes i) smooth puree, ii) lumpy mash, iii) chewable food and iv) drink. Then let the child complete the snack with what ever food/drink is preferable.
   - The spoon should be presented at mouth level approximately 10 cms from the face. Allow the child to see the approaching spoon and respond.
   - Continue to videotape the entire snack.

4. Post-feeding Check:
   - **'Wet/gurgly breathing'** item: Repeat procedure as for pre-feeding check. Mark the 'wet/gurgly breathing' item on snack evaluation sheet.
   - **'Wet/gurgly voice'** item: Repeat procedure as for pre-feeding check. Mark the 'wet/gurgly voice' item on the snack evaluation sheet.
   - **'Cough'** item: If the child has been observed to cough at any time since commencing ingestion of the snack (food or fluid), mark the

item as cough observed. If possible mark whether this occurred on foods (puree, lumpy mashed, chewable) or drink or both. Mark the 'cough' items on the snack evaluation sheet.

- **'Drooling'** item. Repeat procedure as for pre-feeding check. Mark the drooling severity score on the snack evaluation sheet.

5. Finish videotape.

6. Data:
    - Put the videotape and Feeding Evaluation Data Sheet into a plastic zip lock bag for analysis by the speech pathologist.
    - Ensure the videotape and Data sheets both have the child's participant number on it.

QLD CP Child: Growth, Nutrition and Physical Activity: Snack Evaluation Data Sheet	Participant ID

FORM TO BE COMPLETED FOR ALL CHILDREN (including non-oral)

Date of Evaluation: ☐☐ / ☐☐ / ☐☐☐☐
Chronological Age: ☐☐ months
Corrected Age: ☐☐ months

Occlusion:
Body position:
Head position:
Head support:

Foods that my child will eat today include:

	Name food/ fluid	Omitted	Reason for Omission
☐ Puree			☐ Safety or aspiration risk ☐ Refusal, but generally eats texture ☐ Refusal, but normally not part of diet
☐ Lumpy Mash			☐ Safety or aspiration risk ☐ Refusal, but generally eats texture ☐ Refusal, but normally not part of diet
☐ Cheese stick			☐ Safety or aspiration risk ☐ Allergies ☐ Refusal, but generally eats texture ☐ Refusal, but normally not part of diet
☐ Arrowroot biscuit			☐ Safety or aspiration risk ☐ Allergies ☐ Refusal, but generally eats texture ☐ Refusal, but normally not part of diet
FLUIDS: Include <u>either</u> thin or thick fluids			
☐ Thin Drink cup			☐ Safety or aspiration risk ☐ Refusal, but generally drinks with cup ☐ Refusal, but utensil not normally used
☐ Thin Drink straw			☐ Safety or aspiration risk ☐ Refusal, but generally drinks with cup ☐ Refusal, but utensil not normally used
Optional: ☐ Thin Drink other	☐ Bottle ☐ Trainer cup ☐ Pop-top		☐ Safety or aspiration risk ☐ Refusal, but generally drinks with cup ☐ Refusal, but utensil not normally used
☐ Thick Drink cup			☐ Safety or aspiration risk ☐ Refusal, but generally drinks with cup ☐ Refusal, but utensil not normally used
☐ Thick Drink straw			☐ Safety or aspiration risk ☐ Refusal, but generally drinks with cup ☐ Refusal, but utensil not normally used
Optional: ☐ Thick Drink other	☐ Bottle ☐ Trainer cup ☐ Pop-top		☐ Safety or aspiration risk ☐ Refusal, but generally drinks with cup ☐ Refusal, but utensil not normally used

NB. Circle the utensil that is most commonly used at home.

Does the child have a cold/ respiratory infection today? ☐ Yes ☐ No
Compared to when they are well, does their breathing/ chest sound ☐ Better? ☐ Worse?

Chest Status & Clinical Signs of Aspiration:

Clinical Sign	Pre-Feeding	Post-Feeding
1. Wet / gurgly breathing [sound]		
2. Rattly chest [feel]		
3. Wet / gurgly voice [sound]		
4. Cough [visual/sound]		
5. Drooling [visual]	1 2 3 4 5	1 2 3 4 5

For items 1-4 indicate "present" = "1" or "absent" = 0.
Item 5: 1=no loss, 2=lips wet, 3=lips and chin wet, 4=clothes and objects wet, 5=clothes, hands, objects very wet.

SOMA OMC Category: BOTTLE

Name:	Date of Assessment:	Age:

Bottle

Indicate liquid administered:

Non-rateable			Rateable	
Refused	Omitted	Not observed	**Yes**	**No**
React 2	Anticipatory mouth opening		**Y**	**N**
React 4	No liquid enters mouth		**Y**	**N**
Accept 2	Accepts liquid within two seconds		**Y**	**N**
Lip 3	Upper lip firmly seals around teat		**Y**	**N**
Lip 5	Intermittent / incomplete upper lip contact / seal		**Y**	**N**
Lip 6	Intermittent / incomplete upper lip contact / seal		**Y**	**N**
Lip 7	Lip closure during swallow		**Y**	**N**
Jaw 1	Small vertical movements		**Y**	**N**
Sequence 1	Smooth rhythmic sequence		**Y**	**N**
		Sum of shaded boxes		

Cutting Score: **≥ 5 indicates oral motor dysfunction**
< 5 indicates normal oral motor function

SOMA: BOTTLE

Clinical Signs of Aspiration / Pharyngeal Dysfunction	Yes	No
Gagging	**Y**	**N**
Coughing	**Y**	**N**
Choking	**Y**	**N**
Throat Clearing	**Y**	**N**
Vomiting with feeds	**Y**	**N**
Multiple swallows	**Y**	**N**
Wheeze	**Y**	**N**
Stridor (Harsh, high-pitched, vibratory noise) Inspiratory / Expiratory / Biphasic	**Y**	**N**
Increased respiratory rate	**Y**	**N**
Laboured breathing	**Y**	**N**
Wet breathing	**Y**	**N**
Nasal regurgitation during / nasal congestion after feeds	**Y**	**N**
Wet vocalisations / 'gurgly voice'	**Y**	**N**
Runny eyes or 'eye tearing'	**Y**	**N**
Colour changes: circumoral cyanosis or pale/dusky after feeds	**Y**	**N**
Temperature spikes	**Y**	**N**
Refusal to take texture or struggle behaviours throughout feed	**Y**	**N**

Notes:

SOMA OMC Category: TRAINER CUP

Name:	Date of Assessment:	Age:

Trainer Cup

Indicate liquid administered

Non-rateable			Rateable	
Refused	Omitted	Not observed	**Yes**	**No**
Liquid loss	Profuse/marked liquid loss		**Y**	**N**
Sequence 2	Panic reactions when liquid presented		**Y**	**N**
Sequence 3	Choking		**Y**	**N**
Tongue 10	Tongue thrust		**Y**	**N**
Tongue 11	Asymmetry		**Y**	**N**
Jaw 1	Small vertical movements		**Y**	**N**
Jaw 6	Jaw alignment during drinking		**Y**	**N**
Jaw 10	External jaw stabilisation required 100%		**Y**	**N**
Jaw 12	Internal stabilisation		**Y**	**N**
Swallow 1	Jaw alignment		**Y**	**N**
Swallow 4	Panic reaction during / after swallow		**Y**	**N**
Swallow 5	No swallow observed		**Y**	**N**
Swallow 6	Uses gravity (eg. head extension)		**Y**	**N**
Swallow 7	Numerous attempts to initiate swallow		**Y**	**N**
		Sum of shaded boxes		
Cutting Score:	**≥ 5 indicates oral motor dysfunction** **< 5 indicates normal oral motor function**			

Clinical Signs of Aspiration / Pharyngeal Dysfunction	Yes	No
Gagging	**Y**	**N**
Coughing	**Y**	**N**
Choking	**Y**	**N**
Throat Clearing	**Y**	**N**
Vomiting with feeds	**Y**	**N**
Multiple swallows	**Y**	**N**
Wheeze	**Y**	**N**
Stridor (Harsh, high-pitched, vibratory noise) Inspiratory / Expiratory / Biphasic	**Y**	**N**
Increased respiratory rate	**Y**	**N**
Laboured breathing	**Y**	**N**
Wet breathing	**Y**	**N**
Nasal regurgitation during / nasal congestion after feeds	**Y**	**N**
Wet vocalisations / 'gurgly voice'	**Y**	**N**
Runny eyes or 'eye tearing'	**Y**	**N**
Colour changes: circumoral cyanosis or pale/dusky after feeds	**Y**	**N**
Temperature spikes	**Y**	**N**
Refusal to take texture or struggle behaviours throughout feed	**Y**	**N**

SOMA: TRAINER CUP

SOMA OMC Category: CUP

Name:	**Date of Assessment:**	**Age:**

Cup

Indicate liquid administered

Non-rateable			**Rateable**	
Refused	Omitted	Not observed	Yes	No

Accept 2	Accepts within two seconds	**Y**	**N**
Sequence 2	Panic reactions when liquid placed in mouth	**Y**	**N**
Sequence 3	Choking	**Y**	**N**
Liquid Loss	Profuse / marked liquid loss	**Y**	**N**
Tongue 10	Tongue thrust	**Y**	**N**
Tongue 11	Asymmetry	**Y**	**N**
Jaw 1	Small vertical movements	**Y**	**N**
Jaw 4	Jaw clenching	**Y**	**N**
Swallow 9	Gagging	**Y**	**N**
	Sum of shaded boxes		

Cutting Score: **>/= 5 indicates oral motor dysfunction**

< 5 indicates normal oral motor function

SOMA – CUP

Clinical Signs of Aspiration / Pharyngeal Dysfunction	**Yes**	**No**
Gagging	**Y**	**N**
Coughing	**Y**	**N**
Choking	**Y**	**N**
Throat Clearing	**Y**	**N**
Vomiting with feeds	**Y**	**N**
Multiple swallows	**Y**	**N**
Wheeze	**Y**	**N**
Stridor (Harsh, high-pitched, vibratory noise) Inspiratory / Expiratory / Biphasic	**Y**	**N**
Increased respiratory rate	**Y**	**N**
Laboured breathing	**Y**	**N**
Wet breathing	**Y**	**N**
Nasal regurgitation during / nasal congestion after feeds	**Y**	**N**
Wet vocalisations / 'gurgly voice'	**Y**	**N**
Runny eyes or 'eye tearing'	**Y**	**N**
Colour changes: circumoral cyanosis or pale/dusky after feeds	**Y**	**N**
Temperature spikes	**Y**	**N**
Refusal to take texture or struggle behaviours throughout feed	**Y**	**N**

Notes:

SOMA OMC Category: PUREE

Name:	Date of Assessment:	Age:

Puree

Fromage Frais Mousse Pureed Fruit Other (Circle choice)

Non-rateable			Rateable	
Refused	Omitted	Not observed	**Yes**	**No**
React 1	Head orientation to spoon		**Y**	**N**
Sequence 1	Smooth rhythmic sequence		**Y**	**N**
Lip 1	Lower lip draws inwards around spoon		**Y**	**N**
Lip 2	Upper lip removes food from spoon		**Y**	**N**
Lip 3	Lower / upper lip assists in cleaning		**Y**	**N**
Lip 11	Lower lip active during suck / munch / chew		**Y**	**N**
Tongue 11	Consistent / considerable protrusion		**Y**	**N**
Tongue 12	Protrusion beyond incisors		**Y**	**N**
Jaw 1	Graded jaw opening		**Y**	**N**
		Sum of shaded boxes		
Cutting Score:	**>/= 3 indicates oral motor dysfunction** **< 3 indicates normal oral motor function**			

Clinical Signs of Aspiration / Pharyngeal Dysfunction	Yes	No
Gagging	Y	N
Coughing	Y	N
Choking	Y	N
Throat Clearing	Y	N
Vomiting with feeds	Y	N
Multiple swallows	Y	N
Wheeze	Y	N
Stridor (Harsh, high-pitched, vibratory noise) Inspiratory / Expiratory / Biphasic	Y	N
Increased respiratory rate	Y	N
Laboured breathing	Y	N
Wet breathing	Y	N
Nasal regurgitation during / nasal congestion after feeds	Y	N
Wet vocalisations / 'gurgly voice'	Y	N
Runny eyes or 'eye tearing'	Y	N
Colour changes: circumoral cyanosis or pale/dusky after feeds	Y	N
Temperature spikes	Y	N
Refusal to take texture or struggle behaviours throughout feed	Y	N

Notes:

SOMA: PUREE

SOMA OMC Category: SEMI-SOLIDS

Name:	**Date of Assessment:**	**Age:**
Peas Baked Beans Cottage Cheese Other		(Circle choice)

		Non-rateable			**Rateable**	
Refused	Omitted	Not observed			**Yes**	**No**
Drool 1	Consistent / considerable drooling				**Y**	**N**
Sequence 1	Smooth rhythmic sequence				**Y**	**N**
Initiation 1	Sequence initiation within two seconds				**Y**	**N**
Lip 13	Lips closed during swallow				**Y**	**N**
Jaw 1	Graded jaw opening				**Y**	**N**
Jaw 2	Internal jaw stabilisation				**Y**	**N**
Jaw 3	External jaw stabilisation required 100%				**Y**	**N**
Jaw 10	Associated jaw movements				**Y**	**N**
				Sum of shaded boxes		

Cutting Score: **≥ 4 indicates oral motor dysfunction**
< 4 indicates normal oral motor function

SOMA: SEMI-SOLIDS

Clinical Signs of Aspiration / Pharyngeal Dysfunction	**Yes**	**No**
Gagging	**Y**	**N**
Coughing	**Y**	**N**
Choking	**Y**	**N**
Throat Clearing	**Y**	**N**
Vomiting with feeds	**Y**	**N**
Multiple swallows	**Y**	**N**
Wheeze	**Y**	**N**
Stridor (Harsh, high-pitched, vibratory noise) Inspiratory / Expiratory / Biphasic	**Y**	**N**
Increased respiratory rate	**Y**	**N**
Laboured breathing	**Y**	**N**
Wet breathing	**Y**	**N**
Nasal regurgitation during / nasal congestion after feeds	**Y**	**N**
Wet vocalisations / 'gurgly voice'	**Y**	**N**
Runny eyes or 'eye tearing'	**Y**	**N**
Colour changes: circumoral cyanosis or pale/dusky after feeds	**Y**	**N**
Temperature spikes	**Y**	**N**
Refusal to take texture or struggle behaviours throughout feed	**Y**	**N**

Notes:

SOMA OMC Category: SOLIDS

Name:	Date of Assessment:	Age:

Solids

Potato Salad Fruit Salad Other (Circle choice)

Non-rateable			Rateable	
Refused	Omitted	Not observed	**Yes**	**No**
Food Loss 1	None / trivial		**Y**	**N**
Drool 1	Consistent / considerable drooling		**Y**	**N**
Sequence 1	Smooth rhythmic sequence		**Y**	**N**
Lip 1	Lower lip draws inwards around spoon		**Y**	**N**
Lip 2	Upper lip removes food from spoon		**Y**	**N**
Lip 4	Lower lip behind upper teeth / sucking		**Y**	**N**
Lip 11	Lower lip active during suck / munch / chew		**Y**	**N**
Tongue 10	Transient / minimal tongue protrusion		**Y**	**N**
Jaw 1	Graded jaw opening		**Y**	**N**
		Sum of shaded boxes		

Cutting Score: **≥ 4 indicates oral motor dysfunction**
< 4 indicates normal oral motor function

SOMA: SOLIDS

Clinical Signs of Aspiration / Pharyngeal Dysfunction	Yes	No
Gagging	**Y**	**N**
Coughing	**Y**	**N**
Choking	**Y**	**N**
Throat Clearing	**Y**	**N**
Vomiting with feeds	**Y**	**N**
Multiple swallows	**Y**	**N**
Wheeze	**Y**	**N**
Stridor (Harsh, high-pitched, vibratory noise) Inspiratory / Expiratory / Biphasic	**Y**	**N**
Increased respiratory rate	**Y**	**N**
Laboured breathing	**Y**	**N**
Wet breathing	**Y**	**N**
Nasal regurgitation during / nasal congestion after feeds	**Y**	**N**
Wet vocalisations / 'gurgly voice'	**Y**	**N**
Runny eyes or 'eye tearing'	**Y**	**N**
Colour changes: circumoral cyanosis or pale/dusky after feeds	**Y**	**N**
Temperature spikes	**Y**	**N**
Refusal to take texture or struggle behaviours throughout feed	**Y**	**N**

Notes:

SOMA OMC Category: CRACKER

Name:	Date of Assessment:	Age:

Non-rateable			Rateable	
Refused	Omitted	Not observed	Yes	No
Food Loss 1	None / trivial		Y	N
Drool 1	Profuse / marked drooling		Y	N
Initiation 1	Sequence initiated within two seconds		Y	N
Lip 4	Lower lip behind upper teeth to suck		Y	N
Lip 7	Lips close around stimulus during bite		Y	N
Lip 9	Lips close intermittently during suck / munch / chew		Y	N
Tongue 10	Transient / minimal tongue protrusion		Y	N
Tongue 11	Considerable / consistent tongue protrusion		Y	N
Tongue 12	Protrusion beyond incisors		Y	N
Tongue 13	Protrusion beyond lips		Y	N
Jaw 2	Internal jaw stabilisation established		Y	N
Jaw 3	Variable stabilisation (not fully established)		Y	N
Jaw 4	External stabilisation required		Y	N
Jaw 5	Vertical movements		Y	N
Jaw 8	Wide vertical excursions		Y	N
Jaw 9	Small vertical excursions		Y	N
Jaw 11	Associated head movements to bite		Y	N
Jaw 12	Uses fingers to transfer food		Y	N
Swallow 9	Gagging		Y	N
Bite 5	Controlled sustained bite		Y	N
Bite 8	Graded jaw opening		Y	N
Bite 12	Mouths cracker only		Y	N
		Sum of shaded boxes		
Cutting Score:	**≥ 9 indicates oral motor dysfunction** **< 9 indicates normal oral motor function**			

Clinical Signs of Aspiration / Pharyngeal Dysfunction	Yes	No
Gagging	Y	N
Coughing	Y	N
Choking	Y	N
Throat Clearing	Y	N
Vomiting with feeds	Y	N
Multiple swallows	Y	N
Wheeze	Y	N
Stridor (Harsh, high-pitched, vibratory noise) Inspiratory / Expiratory / Biphasic	Y	N
Increased respiratory rate	Y	N
Laboured breathing	Y	N
Wet breathing	Y	N
Nasal regurgitation during / nasal congestion after feeds	Y	N
Wet vocalisations / 'gurgly voice'	Y	N
Runny eyes or 'eye tearing'	Y	N
Colour changes: circumoral cyanosis or pale/dusky after feeds	Y	N
Temperature spikes	Y	N
Refusal to take texture or struggle behaviours throughout feed	Y	N

Notes:

SOMA: CRACKER

DYSPHAGIA DISORDER SURVEY – PAEDIATRIC

SCORES	Raw Score	Disability Percentile
PART1. Related Factors	_____	_____
PART 2. Feeding Competency	_____	_____
TOTAL	_____	_____
LEVEL OF EATING AND SWALLOWING COMPETENCY		
1. No Disorder	_____	_____
2. Mild Disorder	_____	_____
3. Moderate Disorder	_____	_____
4. Severe Disorder	_____	_____
5. Profound Disorder	_____	_____

Part 1. RELATED FACTORS

	Item Score
1. BODY MASS INDEX: ___ WNL☐ < NL ☐ <75%NL ☐ HT___ HT/WT: WNL☐ ≤25%ile☐ ≤10%ile☐ WT___	
2. DIET: cut up/whole☐ modified (soft chew, lumpy) ☐ puree ☐ tube ☐ liquid unrestricted ☐ restricted ☐	
3. INDEPENDENCE: self-feeder ☐ assisted self-feeder ☐ dependent feeder ☐ tube feeder ☐	
4. ADAPTIVE UTENSILS USED: none ☐ spoon☐ cup☐ tube☐	
5. POSITIONING: upright independent☐ upright assisted ☐ reclining ☐	
6. POSTURAL CONTROL: trunk stable☐ unstable☐ head/neck stable ☐ unstable☐	
7. FEEDING TECHNIQUES: normal☐ adaptive☐ mal-adaptive☐	

Part 2 Standardised. FEEDING & SWALLOWING COMPETENCY

	Non Chewable	Chewable	Liquid
8. ORIENTING (alerting to food, moving toward food, mouth opening)			
9. RECEPTION (stripping spoon, biting, sipping from cup, appropriate bolus size, timing)			
10. CONTAINMENT (no dribbling or ejecting food or liquid)			
11. ORAL TRANSPORT (no residual in mouth after swallow, efficient bolus transit)			
12. CHEWING (chew adequate bolus, no special placement required)			
13. ORAL-PHARYNGEAL SWALLOW (prompt, sequential liquid swallow, no gagging or multiple swallows)			
14. POST-SWALLOW (absent coughing, wet breath sounds or wet voice) rpt/obs			
15. GASTRO-ESOPHAGEAL FUNCTION (absent vomiting or rumination) rpt/obs			
PART 2 SUB-SCORES			

Dysphagia Severity Scale=

Pre-Speech Assessment Scale

1982 Revised Edition: Suzanne Evans Morris

Summary of Scores:

II Sucking

	Normal scoring range	Abnormal (-)	Normal (+)
5. Bottle or breast	1-12 months		
6. Cup	6-24+ months		
7. Spoon	3-24+ months		
	TOTAL:		
	Average:		

III Swallowing

	Normal scoring range	Abnormal (-)	Normal (+)
8. Liquids	1-24+ months		
9. Purees	3-24+ months		
10. Semi-solid/ Solids	6-24+ months		
11. Coordination	1-15 months		
	TOTAL:		
	Average:		

IV Biting and Chewing

	Normal scoring range	Abnormal (-)	Normal (+)
13. Jaw/ Biting	5-24 months		
14. Jaw/ Chewing	5-24+ months		
15. Tongue/ Chewing	6-24+ months		
16. Lips/ Chewing	6-24 months		
	TOTAL:		
	Average:		

	PSAS Totals	PSAS Averages
Abnormal (-)		
Normal (+)		

5 SUCKING: LIQUIDS FROM THE BOTTLE OR BREAST

SCORE	BEHAVIOURAL DESCRIPTION	
-9	Enlarged nipple hole (liquid flows automatically into the mouth for swallowing). No or minimal sucking motions are observed. Sucking may be interfered with by jaw thrust, tongue thrust, tongue retraction, biting or mouthing, hypertonia or hypotonia of the tongue which limit tongue movement. OR	pom_btnp pom_btnosk pom_btab
	Liquids are provided by tube feedings or spoon.	pom_bttb pom_btspn
-6	Sucking or suckling motions occur intermittently/ throughout feeding but interfered with by chewing or biting of the nipple, tongue thrust, tongue retraction, or jaw thrust.	pom_btab
	Hard approximation of the tongue with the palate makes insertion of the nipple difficult (but with effort the nipple is inserted and some sucking or suckling is observed).	pom_btins
-4	The nipple hole is enlarged, but child sucks/ suckles nipple without biting, chewing, tongue thrust, tongue retraction or jaw thrust.	pom_btnp
	Sucking is slow, inefficient, weak or poorly sustained which may result in a small intake of liquid (ie 50mL or less in 30 minutes).	pom_btinef
-3	Exaggerated tongue protrusion during sucking or as the nipple is inserted or removed. A true tongue thrust is not observed.	pom_bttpr
	A normal cupped configuration of the tongue is never observed during sucking or as the nipple is removed or reinserted. The tongue is passively flat, bunched, or humped during sucking.	pom_btntcp
-1	Sucking patterns show mildly abnormal components which may or may not have been previously described. Or the abnormal components occur with extremely low frequency or only under stress or illness.	pom_btmdab
X	Lack of bottle or breast feeding is not based on the presence of abnormal oral movements which would interfere with the sucking process.	
1 month	Uses a suckling or sucking pattern. May lose liquid during sucking/ swallowing or as the nipple is inserted or removed.	pom_btsk pom_btll
6 months	Uses a suckling or sucking pattern. Does not lose liquid during sucking/ swallowing. Slight loss as the nipple is inserted or removed.	pom_btsk pom_btllins
9 months	Uses a suckling or sucking pattern. Does not lose liquid.	pom_btsk pom_btnoll
12 months	The child is chronologically older than 12 months and takes liquids from cup or spoon. May continue with the bottle in the evening before going to sleep.	

6 SUCKING: LIQUIDS FROM THE CUP

SCORE	BEHAVIOURAL DESCRIPTION	
-9	No or minimal sucking movement occurs when liquid is placed in the mouth. Liquid is poured into the mouth from the cup with no initiating suck.	pom_cpnosk
-8	Sucking or suckling is present during cup drinking. Abnormal oral patterns **>50%**. This interference is caused by: a) Jaw thrust, jaw clenching, tonic bite reflex, phasic bite reflex, chewing or uncoordinated mouthing of the cup or liquid in the mouth. b) Tongue thrust, strong retraction of the tongue, or hard approximation of the tongue with the palate. c) Lip retraction or purse-string action. d) Severe hypotonia or muscle weakness of tongue, lips or jaw. e) Sucking or suckling movements which are abnormally slow or unsustained. f) Lack of an initiating suck. Sucking begins only after liquid is poured in.	pom_cpskl pom_cpab51
-6	Sucking or suckling is present during cup drinking. Abnormal oral patterns **<50%**. This interference is caused by: a) Jaw thrust, jaw clenching, tonic bite reflex, phasic bite reflex, chewing or uncoordinated mouthing of the cup or liquid in the mouth. b) Tongue thrust, strong retraction of the tongue, or hard approximation of the tongue with the palate. c) Lip retraction or purse-string action. d) Severe hypotonia or muscle weakness of tongue, lips or jaw. e) Sucking or suckling movements which are abnormally slow or unsustained. Lack of an initiating suck. Sucking begins only after liquid is poured in.	pom_cpsk pom_cpab49
-3	Exaggerated tongue protrusion. A true tongue thrust is not observed.	pom_cptpr
	Normal cupped configuration of tongue is never observed in drinking or as cup is inserted or removed. Tongue is passively flat, bunched or humped in drinking.	pom_cpntcp
-1	Sucking patterns show mildly abnormal components which may or may not have been previously described or occur with extremely low frequency or only under stress or illness.	pom_cpmdab
O or X	Child is chronologically under 6 months and does not take liquids from the cup. Or cup drinking has not been introduced and/ or tested because of reasons not related to a primary sucking or swallowing disorder (gastro-intestinal disorders, structural disorders, delayed oral development due to prematurity, mental retardation, exclusive use of breast feeding, or other medical or behavioural).	
6 months	Child uses primarily a **suckling** pattern or a mixture of sucking and suckling. Extension-retraction motion of tongue. Jaw movement is wide up-down or backward-forward motion. Loses liquid.	pom_cpskl pom_cpexr pom_cpjwd pom_cpll
12 months	Uses a **sucking** pattern. **Extension-retraction of tongue not observed**. Jaw movement may be in a wide up-down or backward-forward direction. Tongue may protrude beneath cup. May lose liquid.	pom_cpsk pom_cpnoexr pom_cpjwd pom_cptuc pom_cpll
18 months	Uses a sucking pattern. **External jaw stabilization** is obtained by biting down on edge of cup. Upper lip is closed on edge of cup. Tongue does not protrude from mouth or rest beneath cup. May lose liquid.	pom_cpsk pom_cpexjst pom_cpulcl pom_cpntuc pom_cpll
	Minimal up-down or backward-forward movement: child moving gradually from an unstabilized jaw movement toward internal jaw stabilization.	pom_cpinjst
24 months	Uses a sucking pattern. **Internal jaw stabilization <75%** during consecutive sips, obtained through co-activation of the jaw opening and closing muscles. Pattern may alternate with slight up-down motions or biting on cup. May lose liquid.	pom_cpsk pom_cpinjst74 pom_cpll
24+ months	Uses a sucking pattern. **Active internal jaw stabilization >75%** during consecutive sips, without biting on cup. Pattern may alternate with slight up-down motion or biting on cup. May lose liquid.	pom_cpsk pom_cpinjst76 pom_cpll

8 SWALLOWING: LIQUIDS

SCORE	BEHAVIOURAL DESCRIPTION	
-9	Child takes no liquids by mouth due to a primary sucking or swallowing disorder.	
	Child is on **tube feeding for liquids or takes thickened liquid in from spoon**.	pom_bttb
	No swallowing movements are observed when liquid reaches the back of the mouth.	pom_btspn
-8	**Swallowing movements occur spontaneously** during feeding, but the child does not take enough liquid by mouth to prevent dehydration. **Additional liquid is given by tube feeding.**	pom_swlsptb
	Strong head extension or flexion during swallowing of liquids is observed more than 75% of the time. The child resists positioning to maintain the head upright for swallowing.	pom_swlhdpsn
-6	**All liquids are given by mouth** and supplemental tube feedings are not given.	pom_swltrol
	Liquids may be thickened to increase the ease of swallowing: however, a liquid consistency is maintained. Swallowing is considered poor or difficult because of:	pom_swltkorl
	a) Nasal regurgitation.	pom_swlnsl
	b) Very slow swallowing due to difficulty moving the liquid backward into the pharynx for swallowing, or pooling of liquid in the oro-pharynx.	pom_swlott
	c) Very passive swallowing with minimal upward movement of larynx and hyoid bone during the swallow.	pom_swlps
	d) Spontaneous use of head extension to assist with the swallow: doesn't resist attempts to reduce head extension.	pom_swlhdpsn
	e) Lip, tongue or jaw retraction. Jaw thrust, or severe tongue thrust which appears to interfere with swallowing.	pom_swlorlab
	f) Coughing, choking, or gagging, occurring more than 3 times during a meal.	pom_swlph
	g) Greater difficulties with certain types of liquid (i.e., thin, milky).	pom_swltxt
-4	**Tongue thrust** is observed during swallowing which may or may not interfere with the movement of liquid into the pharynx for efficient swallowing.	pom_swltth
-3	**Exaggerated tongue protrusion** is observed during swallowing. A true tongue thrust is not observed.	pom_swltpr
-1	Swallowing patterns show mildly abnormal components which may or may not have been previously described. Or the abnormal components occur with extremely low frequency or only under stress or illness.	pom_swlmdab
X	The child does not take liquids but this is unrelated to a primary sucking or swallowing disorder (eg. gastro-intestinal disorders, structural disorders, delayed oral development due to prematurity or other medical or behavioral reasons).	
1 month	Swallows thin liquids from the **bottle or breast**.	pom_swlbt
	Tongue may protrude with an extension-retraction movement pattern during the swallow or it may simply protrude between the teeth.	pom_swlexr
6 months	**Swallows liquid from the cup** with no observable elevated tongue tip position.	pom_swlcpnet
	Tongue protrudes with an extension-retraction movement pattern during the swallow or shows a simple protrusion between the teeth.	pom_swlexr
	Lips may be open during the swallow.	pom_swllop
	There may be loss of liquid.	pom_swlll
12 months	Swallows liquid from the cup with an **intermittent elevated tongue-tip position**. This pattern may alternate with either an extension-retraction pattern or simple protrusion of the tongue between the teeth.	pom_swlcpet10
	Lips may be open during the swallow.	pom_swllop
	There may be loss of liquid.	pom_swlll
24 months	Swallows from the cup with an **elevated tongue position is used intermittently or consistently** for swallowing.	pom_swlqet100
	Easy lip closure.	pom_swllcl
	No loss of liquid both during drinking and after the cup is removed from the mouth.	pom_swlnll
24+ months	Swallows with **no observable extension-retraction or protrusive movements** of the tongue.	pom_swlnexr
	Easy lip closure	pom_swllcl
	No liquid loss during drinking or after the cup is removed from the mouth.	pom_swlnll

11 COORDINATION OF SUCKING, SWALLOWING AND BREATHING

SCORE	BEHAVIOURAL DESCRIPTION	
-1	The coordination of sucking, swallowing and breathing shows mildly abnormal components which may or may not have been previously described. Or the abnormal components occur with extremely low frequency or only under stress or illness.	pom_swcdnmdab
X	The child does not take liquids unrelated to a primary sucking, swallowing or respiratory disorder (due to gastro-intestinal disorders, structural disorders, delayed oral development due to prematurity or other medical or behavioural reasons).	
1 month	Child sequences **2 or more sucks** from the bottle or breast before pausing to breathe or swallow.	pom_swcdn2skb
	Breathing may become noisier doing feeding.	pom_swcdnnsy
3 months	Long sequences of **twenty or more sucks** are present with the bottle.	pom_swcdn20
	Swallowing follows sucking with **no discernable pauses** when the child is hungry and not looking around. Pauses for breathing are infrequent. Sucking motions occur almost simultaneously with swallowing (overlapping motions).	pom_swcdnnps
	Occasional coughing or choking indicating poor timing of the suck-swallow pattern with breathing.	pom_swcdncgh3
6 months	**Long sequences** of sucking-swallowing-breathing are observed with the bottle.	pom_swcdn21
	During cup drinking **many continuous sucks** are observed which are followed by uncoordinated swallowing.	pom_swcdncp3u
	Much liquid is lost from cup.	pom_swcdncpll
	Coughing and choking may result **from intake of larger mouthfuls** of liquid from cup.	pom_swcdncplg
9 months	Long sequences of continuous sucks which are not timed with swallowing may continue to occur.	pom_swcdncp3u
	During cup drinking the child takes **1-3 sucks before stopping/ pulling away to swallow or breathe.**	pom_swcdncp3p
	Coughing, choking or sputtering may occur.	pom_swcdncgh
12 months	Swallowing follows sucking with no pause as the child drinks from the cup.	pom_swcdncpnps
	Sequences 3+ suck-swallows when the child is thirsty.	pom_swcdncp3c
	Intake during consecutive suck-swallows is **less than 30mL**.	pom_swcdn29ml
	Some coughing and choking may continue to occur.	pom_swcdncgh
15 months	Swallowing follows sucking with no pause as the child drinks from the cup.	pom_swcdncpnps
	Sequences 3+ suck-swallows while drinking.	pom_swcdncp3c
	Shorter suck-swallow sequences may continue when not thirsty or interested in drinking.	
	Intake during consecutive suck-swallows is **30mL or more** with no major pauses.	pom_swcdn31ml
	Coughing and choking are rarely observed (pattern is well-coordinated with respiration).	pom_swcdnncgh

7 SUCKING: PUREED FOOD FROM A SPOON

SCORE	BEHAVIOURAL DESCRIPTION	
-9	No or minimal sucking movement occurs when pureed food is placed in the mouth. Food may be poured in from the spoon. The tongue moves food uncoordinatedly, and some may be swallowed.	pom_prnosk
-8	Sucking or suckling. Abnormal oral patterns **>50%** of the time when the spoon is presented or enters the mouth, or food is being sucked to the back of the mouth. Interference is caused by: a) Jaw thrust, jaw clenching, tonic bite reflex, phasic bite reflex, chewing or uncoordinated mouthing of the food in the mouth. b) Tongue thrust, strong retraction of the tongue, or hard approximation of the tongue with the palate. c) Lip retraction or purse-string action. d) Severe hypotonia of tongue, lips or jaw. e) Sucking or suckling movements which are abnormally slow or unsustained.	pom_prsk pom_prab51
-6	Sucking or suckling. Abnormal oral patterns **< 50%** of the time when the spoon is presented or enters the mouth, or food is being sucked to the back of the mouth. Interference is caused by: a) Jaw thrust, jaw clenching, tonic bite reflex, phasic bite reflex, chewing or uncoordinated mouthing of the food in the mouth. b) Tongue thrust, strong retraction of the tongue, or hard approximation of the tongue with the palate. c) Lip retraction or purse-string action. d) Severe hypotonia of tongue, lips or jaw. Sucking or suckling movements which are abnormally slow or unsustained.	pom_prsk pom_prab49
-3	Exaggerated tongue protrusion is observed during sucking or as the spoon is inserted or removed. A true tongue thrust is not observed.	pom_prtpr
	A normal cupped configuration of the tongue is never observed when the spoon is presented, enters the mouth or as the food is being sucked to the back of the mouth. The tongue is passively flat, bunched or humped.	pom_prntcp
-1	Sucking patterns show mildly abnormal components which may or may not have been previously described. Or the abnormal components occur with extremely low frequency or only under stress or illness.	pom_prmdab
O or X	Child is chronologically under 3 months and does not take food from spoon. Or cup drinking has not been introduced and/ or tested because of reasons not related to a primary sucking or swallowing disorder (gastro-intestinal disorders, structural disorders, delayed oral development due to prematurity, mental retardation, exclusive use of breast feeding, or other medical or behavioural).	
3 month	**Suckling or sucking** pattern is observed in the tongue and/or jaw as food approaches mouth or touches lips. **Upper lip does not assist** in removal of food from spoon.	pom_prskl
6 months	The child shows visual or tactile recognition of the spoon. The **tongue and jaw remain quiet** until the food enters the mouth. **Upper lip is slightly forward or downward** but does not show a downward and forward movement which actively cleans the spoon.	pom_pror pom_prqt pom_prnul
8 months	Upper lip moves downward and forward to posture or rest on the spoon and assist in removing food from the spoon.	pom_prulcl
10 months	Upper lip actively removes the food from the spoon. **Lower lip** draws inward as the spoon is removed or as food remains on the lower lip. Specific cleaning movements are not observed.	pom_prulcl pom_prlwl pom_prncln
15 months	**Upper incisors are used to clean** the lower lip as it draws inward. Tongue show sucking or a mixture of **sucking and suckling**. **Phasic bite reflex is not present** at any time; however, some playful biting on the spoon in a game-like fashion may continue to occur.	pom_pruicln pom_prskl50 pom_prnphb
24+ months	**Tongue is used to clean** food from the upper/ lower lips (free sweeping movement). Tongue elevation and depression are **independent** of jaw movement and show some skilful action of the tongue tip. Slight lateral movements of the jaw may be observed. **Intermittent suckling** movements of the tongue may occur.	pom_prtcln pom_prtind pom_prltjw pom_prskl10

9 SWALLOWING: PUREES

SCORE	BEHAVIOURAL DESCRIPTION	
-9	Child does not take purees by mouth due to a primary sucking or swallowing disorder. Child may be tube fed. No swallowing movements are observed when purees are placed in the mouth.	pom_swprtb
-8	Swallowing movements occur spontaneously during feeding, but the child doesn't take enough food by mouth **(supplemental tube feeds).**	pom_swprsptb
	Strong head extension or flexion during swallowing of semi-solids is observed >75% of the time. The child resists positioning to maintain the head upright for swallowing.	pom_swprhdpsn
-6	All purees are given by mouth and **tube feedings are not given to supplement food intake** (may receive tube feedings for liquids). Swallowing of semi-solids is considered poor or difficult because of:	
	a) Nasal regurgitation.	pom_swprnsl
	b) Very slow swallowing due to extreme difficulty moving the food backward into the pharynx for swallowing, or pooling of food in the oro-pharynx.	pom_swprott
	c) Very passive swallowing with minimal upward movement of the larynx and hyoid bone during the swallow.	pom_swprps
	d) Spontaneous use of head extension to assist with the swallow. Doesn't resist attempts to reduce head extension.	pom_swprhdpsn
	e) Lip, tongue or jaw retraction, jaw thrust, or severe tongue thrust which appears to interfere with swallowing.	pom_swprorlab
	f) Gags, chokes, coughs, vomits, or spits semi-solid foods more than 25% of the time.	pom_swprph
-4	**Tongue thrust** is observed during swallowing which may or may not interfere with movement of food into the pharynx for efficient swallowing.	pom_swprtth
-3	**Exaggerated tongue protrusion** observed during swallowing. A true tongue thrust is not observed.	pom_swprtpr
	The child is able to swallow purees only when excessive suckling action, the thumb, a pacifier or other **object is placed in the mouth to trigger a suck-swallow sequence** >50% of the time.	pom_swprprp
-1	Swallowing patterns show mildly abnormal components which may or may not have been previously described. Or the abnormal components occur with extremely low frequency or only under stress or illness.	pom_swprmdab
0 or X	The child does not take purees unrelated to a primary sucking or swallowing disorder (due to age, exclusive breast feeding, gastro-intestinal disorders, delayed oral development due to prematurity or other medical or behavioral reasons).	
3 months	Swallows soft or pureed foods (semi-solids).	
	Gagging, choking, coughing, vomiting or spitting occur **<25%** of the time.	pom_swprph24
	Primitive **suckle-swallow** response to move food into the pharynx for swallowing.	pom_swprskl
	Some food is pushed out of the mouth.	pom_swprflpsh
6 months	Gagging, choking, coughing, vomiting, or spitting occur **less than 3 times/ meal**.	pom_swprph3
	Tongue shows an **extension-retraction pattern or simple protrusion** between the teeth during the swallow.	pom_swprexr
	Minor loss of food/ saliva (food is not pushed out of the mouth by the tongue).	pom_swprfl
9 months	**Sucking pattern** (up-down tongue movements) with intermittent suckle (tongue extension-retraction) to move food into the pharynx for swallowing.	pom_swprsk
	Tongue shows a simple protrusion between the teeth or gums.	pom_swprtpr
12 months	**Intermittent elevated tongue-tip position**, which may alternate with tongue protrusion.	pom_swpret10
	Easy lip closure.	pom_swprlcl
	No loss of food or saliva.	pom_swprnfl
18 months	Elevated tongue position is used intermittently or consistently for swallowing.	pom_swpret100
	Some simple protrusion of the tongue may be observed during swallowing.	pom_swprtpr
	No extension-retraction movements of the tongue are present.	pom_swprnexr
24+ months	An elevated tongue is used for swallowing.	pom_swpret100
	No tongue protrusion is observed.	pom_swprntpr
	Swallows with no loss of food or saliva.	pom_swprnfl

III BITING AND CHEWING

13 JAW MOVEMENT IN BITING

SCORE	BEHAVIOURAL DESCRIPTION	
-9	Child shows no biting of food because of interference by abnormal movement patterns of the jaw such as jaw thrust, jaw clenching, excessive jaw retraction or tonic bite reflex or severe hypotonicity or flaccidity making active jaw closure impossible.	pom_jbnb
-8	Biting occurs when food is presented between the gums or teeth. Abnormal movement patterns of the jaw occur **>50%** of the time when food is presented. This interference is caused by:	pom_jbab51
	a) Excessive jaw retraction,	pom_jbjwr
	b) Jaw thrust.	pom_jbjwth
	c) Tonic bite reflex.	pom_jbtbr
	d) Involuntary movement or tremor.	pom_jbim
	e) Marked hypotonicity or muscle weakness.	pom_jbhypo
	f) Jaw clenching or tooth grinding.	pom_jbjwcl
-6	Some biting occurs when food is presented for biting. Abnormal movement patterns of the jaw occur **<50%** of the time when food is presented. Interference is caused by:	pom_jbab49
	a) Excessive jaw retraction,	pom_jbjwr
	b) Jaw thrust.	pom_jbjwth
	c) Tonic bite reflex.	pom_jbtbr
	d) Involuntary movement or tremor.	pom_jbim
	e) Marked hypotonicity or muscle weakness.	pom_jbhypo
	f) Jaw clenching or tooth grinding.	pom_jbjwcl
-3	The child shows **difficulty biting through soft or hard foods** due to mild hypotonicity or lack of power in the jaws or inability to stabilize the jaw long enough to bite through.	pom_jbnsb
-1	Jaw movement patterns in biting show mildly abnormal components which may or may not have been previously described. Or the abnormal components occur with extremely low frequency or only under stress or illness.	pom_jbmdab
0 or X	The child is under 5 months of age or does not make any attempt to bite the food. Sucking, suckling, or a complete lack of response is observed. However, there are not abnormal oral patterns of the jaw which would interfere with the biting process. Abnormal oral patterns of the tongue or lips may be present and may interfere with biting development.	
5 months	Uses a **primitive phasic bite and release pattern** on a softer cookie. There is a relatively regular biting rhythm and lack of a sustained controlled bite through the cookie.	pom_jbphb
	Pieces of cookie may come off and the child may occasionally use a sucking **or suckling pattern** instead of an attempted bite.	pom_jbskl
9 months	Holds the soft cookie **between the gums or teeth without biting through**.	pom_jbqtjw
	Maintains a quiet jaw and holding posture as feeder assists in breaking off a piece.	pom_jbcrbk
	May revert to a primitive phasic bite pattern or sucking.	pom_jbphb
12 months	Uses a **controlled, sustained bite on a soft cookie**.	pom_jbsbsft
	When biting a **hard cookie the bite may be unsustained** because of lack of teeth or biting power -- child may revert to the primitive phasic biting pattern or sucking.	pom_jbnsbhd
18 months	Uses a **controlled, sustained bite on a hard cookie**	pom_jbsbhd
	Presence of overflow or associated movements in the arms or legs, or	pom_jbovflw
	head extension and pulling away to assist with the biting,.	pom_hdbk
21 months	Uses a controlled, sustained bite on a hard cookie	pom_jbsbhd
	N**o overflow or associated movements** in the arms or legs. The head does not extend to assist in biting.	pom_jbnovflw
	May turn head in the direction of the food when presented to the side.	pom_jbtnhd
	Full open mouth position may be used in preparation for biting food of different thicknesses (lacks grading).	pom_jbnjwgd
24 months	Uses a sustained controlled bite	pom_jbsbhd
	Keeps head in midline when food is presented on both sides.	pom_jbhdmd
	Child is able to **grade the opening of the jaw** appropriately when asked to bite food of different thicknesses.	pom_jbjwgd

14 JAW MOVEMENT IN CHEWING

SCORE	BEHAVIOURAL DESCRIPTION	
-9	Child shows no chewing of food.	pom_jcnchw
	There is a contributing influence of abnormal movement patterns of the jaw such as jaw thrust, jaw clenching or tooth grinding, excessive jaw retraction, tonic bite reflex, or severe hypotonicity or flaccidity making active jaw closure or opening impossible.	pom_jcab51
-8	Munching or chewing occurs when solid food is placed in the mouth without biting.	pom_jcab51
	Abnormal movement patterns of the jaw occur **>50%** of the time. This interference is caused by:	
	a) Excessive jaw retraction.	pom_jcjwr
	b) Jaw thrust.	pom_jcjwth
	c) Tonic bite reflex.	pom_jctbr
	d) Involuntary movement or tremor.	pom_jcim
	e) Marked hypotonicity or muscle weakness.	pom_jchypo
	f) Jaw clenching or tooth grinding.	pom_jcjwcl
	g) Excessive jaw protrusion or lateral deviation.	pom_jcjwpr
-6	Munching or chewing occurs when solid food is placed in the mouth without biting.	pom_jcab49
	Abnormal movement patterns of the jaw occur **<50%** of the time. Interference is caused by:	
	a) Excessive jaw retraction.	pom_jcjwr
	b) Jaw thrust.	pom_jcjwth
	c) Tonic bite reflex.	pom_jctbr
	d) Involuntary movement or tremor.	pom_jcim
	e) Marked hypotonicity or muscle weakness.	pom_jchypo
	f) Jaw clenching or tooth grinding.	pom_jcjwcl
	g) Excessive jaw protrusion or lateral deviation.	pom_jcjwpr
-3	The child appears to have lack of power in the jaw or mild hypotonicity in the jaw closing muscles or lack of jaw stability which interferes with the closing-grinding phase of chewing.	pom_jcnsb
-1	Jaw movement patterns in chewing show mildly abnormal components which may or may not have been previously described. Or the abnormal components occur with extremely low frequency or only under stress or illness.	pom_jcmdab
0 or X	The child is under 6 months of age; or the child does not make any attempt to munch or chew the food. Sucking, suckling, or a complete lack of response is observed. However, there are no abnormal movement patterns of the jaw which would interfere with the chewing process. Abnormal movement patterns of the tongue or lips may be present and may interfere with chewing development.	
5 months	Jaw movement in chewing is primarily the primitive **phasic bite-and-release pattern** with a fairly regular stereotyped rhythm.	pom_jcphb
	Diagonal-rotary movements may occur as food is transferred to the side or middle of the mouth.	pom_jcdiagtrf
	Non-stereotyped vertical movements may occur intermittently.	pom_jcvert10
6 months	Jaw movement in chewing is primarily a **non-stereotyped vertical movement** (more variable and less automatic than that described as a phasic bite-and-release pattern).	pom_jcvert80
	Diagonal-rotary movements or phasic bite-and-release movements may occur.	pom_jcdiagtrf
9 months	Jaw movement in chewing is primarily a **non-stereotyped vertical movement**.	pom_jcvert80
	Diagonal-rotary jaw movements occur as food is transferred to the side or middle of the mouth by the tongue.	pom_jcdiagtrf
	The phasic bite-and-release pattern may be observed occasionally if the child is chewing food between the upper and lower central incisors.	pom_jcphbft
15 months	Jaw movement is mixture of **non-stereotyped vertical/ diagonal-rotary** movements.	pom_jcvert50
	Diagonal-rotary movements occur as food is transferred to the sides and to the center.	pom_jcdiagtrf
	Rotary jaw movements are smooth and well coordinated.	pom_jcrty
24+ months	Jaw movement in chewing is primarily a **non-stereotyped vertical movement**.	pom_jcvert80
	Some diagonal-rotary jaw movements are observed.	
	Circular-rotary jaw movements occur as the child transfers food across midline from one side of the mouth to the other.	pom_jccrtytrf

15 TONGUE MOVEMENT IN CHEWING

SCORE	BEHAVIOURAL DESCRIPTION	
-9	Child shows no chewing of food. There is a contributing influence of abnormal movement patterns of the tongue such as tongue thrust, tongue retraction. strong hypersensitivity of the anterior half of the tongue, or hypotonicity of flaccidity of the tongue making tongue movement difficult or impossible.	pom_jcnchw pom_tcab51
-8	Some munching or chewing occurs when solid food is placed in the mouth. Abnormal movement patterns of the tongue occur **> 50%** of the time when food enters the mouth without biting or during chewing. This interference is caused by: a) Tongue retraction. b) Tongue thrust. c) Strong hypersensitivity or the anterior half of the tongue. d) Involuntary movement or tremor. e) Marked hypotonicity or muscle weakness.	pom_tcab51 pom_tctrt pom_tctthr pom_tchsens pom_tcim pom_tchypo
-6	Some munching or chewing occurs when solid food is placed in the mouth. Abnormal movement patterns of the tongue occur **<50%** of the time when food enters the mouth without biting or during chewing. This interference is caused by: a) Tongue retraction. b) Tongue thrust. c) Strong hypersensitivity or the anterior half of the tongue. d) Involuntary movement or tremor. e) Marked hypotonicity or muscle weakness.	pom_tcab49 pom_tctrt pom_tctthr pom_tchsens pom_tcim pom_tchypo
-3	The tongue appears passive or hypotonic, with movement during chewing mildly to moderately reduced.	pom_tcps
-1	Tongue movements in chewing show mildly abnormal components which may or may not have been previously described. Or the abnormal components occur with extremely low frequency or only under stress or illness.	pom_tcmdab
0 or X	The child is under 6 months of age; or the child does not make any attempt to munch or chew the food. Sucking, suckling or a complete lack of response is observed. However, there are no abnormal movement patterns of the tongue which would interfere with the chewing process. Abnormal movement patterns of the jaw or lips may be present and way interfere with chewing development.	
6 months	The tongue shows predominately a munching pattern. No lateralisation of the tongue with solid foods. Sucking or suckling movements may alternate with the munching pattern.	pom_tcmch80 pom_tcnlat pom_tcskl
7 months	Tongue **begins to show some lateralization** with a gross rolling movement or simple horizontal shifts when food is placed between the biting surfaces in the molar area. The tongue is able to move to the side in this manner or may revert to a **suckling pattern when food is placed in the center of the tongue** or needs to be transferred from side to side. **Movement to both sides may not be seen**.	pom_tclatsc
9 months	Lateral movements of the tongue continue when food is placed on the sides Lateral movements of the tongue are **beginning to occur in transferring food from the center to the side**. This may not be seen with high frequency nor to both sides. Intermittent extension-retraction movements may continue in conjunction with a transfer movement which is difficult.	pom_tclatsc pom_tclat1cs20 pom_tcexr10
12 months	When food is placed in the center of the mouth the child is **able to transfer it to both sides with tongue movements**. Intermittent extension-retraction movements may continue in conjunction with a transfer movement which is difficult.	pom_tclat2cs pom_tcexr10
24 months	When food is placed between the biting surfaces of the molars the tongue transfers it to the other side. **Transfer of food across midline** occurs when food is placed on both sides of the mouth. The child may show a preference or greater skill on one side. Movements may be slow or gross. Midline transfers are spontaneous and automatic. Greater difficulty or failure to transfer may occur when the child attempts to transfer volitionally on command. Extension-retraction movements occur intermittently..	pom_tcmdtrf pom_tcslw pom_tcexr10
24+ months	Food can he transferred from **center-to-side and from side-to-side across midline with equal skill.** The child can do this rapidly and with some skill. Speed and skilfulness may not be consistent. He or she may be able to transfer food across midline on command. Precise movements involving the elevated tongue tip may be observed. **Extension-retraction movements do not occur**.	pom_tcmdtrf pom_tcnexr

16 LIP MOVEMENT IN CHEWING

SCORE	BEHAVIOURAL DESCRIPTION	
-9	Child shows no chewing of food. There is a contributing influence of abnormal movement patterns of the lips such as lip retraction, purse-string action, or severe hypotonicity or flaccidity making lip movement impossible.	pom_jcnchw
-8	Munching or chewing occurs when solid food is placed in the mouth. Abnormal movement patterns of the lips occur **>50%** of the time when food is placed in the mouth without biting or during chewing. This interference is caused by:	pom_lcab51
	a) Lip retraction.	pom_lclrt
	b) Lip purse-string action.	pom_lclps
	c) Hypersensitivity with startle as food is on the lips.	pom_lchsens
	d) Involuntary movement or tremor.	pom_lcim
	e) Marked hypotonia or muscle weakness	pom_lchypo
-6	Munching or chewing occurs when solid food is placed in the mouth. Abnormal movement patterns of the lips occur **<50%** of the time when food is placed in the mouth without biting or during chewing. This interference is caused by:	pom_lcab49
	a) Lip retraction.	pom_lclrt
	b) Lip purse-string action.	pom_lclps
	c) Hypersensitivity with startle as food is on the lips.	pom_lchsens
	d) Involuntary movement or tremor.	pom_lcim
	e) Marked hypotonia or muscle weakness	pom_lchypo
-3	Lips remain open during chewing and appear passive or hypotonic with little or no movement, even though the lower jaw moves.	pom_lcps
-1	Lip movement patterns in chewing show mildly abnormal components which may or may not have been previously described. Or the abnormal consonants occur with extremely low frequency or only under stress or illness.	pom_lcmdab
0 or X	The child is under 6 months of age; or the child does not make any attempt to munch or chew the food. Sucking, suckling or a complete lack of response is observed. However, he does not show any abnormal movement patterns of the lips which would interfere with the chewing process. Abnormal movement patterns of the jaw or tongue may be present and may interfere with chewing development.	
6 months	Some munching or chewing patterns.	
	Slight drawing in of either the upper or lower lip or a tightening of the corner of the mouth when food is on the lips.	pom_lclmcln
	Teeth or gums are used to actually clean the food from the lips.	pom_lctcln
9 months	Lips are active with the jaw and **make some mechanical contact** at the sides or in the center as the jaw moves up and down.	pom_lcct
	Upper lip comes forward and down in an active manner during chewing.	pom_lclmchw
	Upper or lower lip draws in when food is on the lips.	pom_lclmcln
12 months	Lips are active in chewing.	pom_lclmchw
	Incisors or gums to clean food from the lower lip as it is drawn inward.	pom_lctcln
	May be loss of food or saliva while chewing.	pom_lcfl
15 months	Upper and lower lips are active in chewing and cleaning.	pom_lclmchw
	Corner of the lip or cheek draws inward and assists in controlling placement or movement of food in the mouth.	pom_lcck
18 months	Lip closure possible during chewing, but intermittent (eg. when he has stuffed his mouth and is in danger of losing food).	pom_lclcl10
	May be some loss of food or saliva while chewing.	pom_lcfl
24 months	Lip closure possible, but may be inconsistent.	pom_lclcl50
	Adequate lip movement during chewing	pom_lclmchw
	No loss of food or saliva from the mouth while chewing.	pom_lcnfl

10 SWALLOWING: SOLIDS

SCORE	BEHAVIOURAL DESCRIPTION	
-9	Child does not take solid foods by mouth due to a primary sucking or swallowing disorder. Child may be tube fed. No swallowing movements are observed when solid foods are placed in the mouth. Active rejection, refusals or inability to swallow solid foods may be related to severe oral hypersensitivity.	pom_swsdno
-8	Strong head extension or flexion during the swallowing of solid foods is observed >75% of the time. The child resists positioning to maintain the head upright for swallowing.	pom_swsdhdpsn
-6	Swallowing of solids is considered poor or difficult because of:	
	a) Nasal regurgitation.	pom_swsdnsl
	b) Very slow swallowing or extreme difficulty moving the food backward into the pharynx for swallowing.	pom_swsdott
	c) Very passive swallowing with minimal upward movement of the larynx and hyoid bone during the swallow.	pom_swsdps
	d) Spontaneous use of head extension to assist with the swallow; doesn't resist attempts to reduce head extension.	pom_swsdhdpsn
	e) Lip, tongue or jaw retraction, jaw thrust, or severe tongue thrust which appears to interfere with swallowing.	pom_swsdorlab
	f) Gags, chokes, coughs, vomits, or spits solid foods more than 25% of the time.	pom_swsdph
	g) Difficulties in forming a bolus for swallowing.	pom_swsdbls
-4	Noticeable **tongue thrust** is observed during swallowing which may or may not interfere with movement of food into the pharynx for efficient swallowing.	pom_swsdtth
-3	Exaggerated tongue protrusion is observed during swallowing. A true tongue thrust is not observed.	pom_swsdtpr
	The child is able to swallow solids only when excessive suckling action, the thumb, a pacifier or other **object is placed in the mouth to trigger a suck-swallow sequence** >50% of the time.	pom_swsdprp
-1	Swallowing patterns show mildly abnormal components which may or may not have been previously described. Or the abnormal components occur with extremely low frequency or only under stress or illness.	pom_swsdmdab
0 or X	The child does not take or swallow solids unrelated to primary sucking or swallowing disorder (due to age, exclusive breast feeding, gastrointestinal disorders, delayed oral development doe to prematurity or retardation, or other medical or behavioral reasons).	
6 months	Swallows some ground, mashed or chopped table foods with noticeable lumps.	pom_swsd
	Gags, chokes, spits, or vomits **<25%** of the time from food of this type contracting or resting on the posterior half of the tongue.	pom_swsdph24
	May use a simple protrusion of the tongue between the teeth or extension-retraction movements.	pom_swsdtpr
12 months	Gagging, choking, vomiting or spitting occur **<3 times/ meal**.	pom_swprph3
	Intermittently elevated tongue-tip position. This pattern may alternate with a simple protrusion of the tongue between the teeth.	pom_swsdet10
	No extension-retraction movements are present during swallowing.	pom_swsdnexr
	May be loss of food or saliva.	pom_swsdfl
18 months	Elevated tongue position is used for swallowing.	pom_swsdet100
	Some protrusive movements of the tongue are observed during swallowing.	pom_swsdtpr
	Easy lip closure as needed	pom_swsdlcl
	No loss of food or saliva.	pom_swsdnfl
24+ months	Elevated tongue position is used for swallowing.	pom_swpret100
	No tongue protrusion is observed during swallowing.	pom_swsdntpr
	Easy lip closure as needed	pom_swprlcl
	No loss of food or saliva.	pom_swprnfl

Appendix 22. Conference Presentations, Invited Speaker and Awards during Candidature

Conference Presentations:

1. Oropharyngeal Dysphagia in Preschool Children with Cerebral Palsy: Comparison between High- and Low-Resource Countries. **Katherine Benfer**, Kelly Weir, Kristie Bell, Robert Ware, Peter Davies, Roslyn Boyd. 68th Annual Meeting of the American Academy of Cerebral Palsy and Developmental Medicine, September 2014.
2. Dietary Intake and Undernutrition in Preschool Children with Cerebral Palsy: Comparison between High- and Low-Resource Countries. **Katherine Benfer**, Kelly Weir, Kristie Bell, Robert Ware, Peter Davies, Roslyn Boyd. 68th Annual Meeting of the American Academy of Cerebral Palsy and Developmental Medicine, September 2014 [poster]
3. Patterns of gross motor severity and motor type in preschool age children with cerebral palsy: comparison between high and low resource countries. **Katherine Benfer**, Rachel Jordan, Sasaka Bandaranayake, Christine Finn, Robert Ware, Roslyn Boyd. 68th Annual Meeting of the American Academy of Cerebral Palsy and Developmental Medicine, September 2014. [presented by Rachel Jordan on behalf of first author]
4. Relationship between brain lesion severity and oropharyngeal dysphagia in young children with cerebral palsy. Kelly Weir, **Katherine Benfer**, Simona Fiori, Kristie Bell, Peter Davies, Roslyn Boyd. 68th Annual Meeting of the American Academy of Cerebral Palsy and Developmental Medicine, September 2014. [poster]
5. Functional oropharyngeal impairments and their relationship to gross motor skills in young children with cerebral palsy. **Katherine Benfer** , Kelly Weir, Kristie Bell, Robert Ware, Peter Davies, Roslyn Boyd. 7th Biennial Conference of the Australasian Academy of Cerebral Palsy and Developmental Medicine, March 2014.
6. Micronutrient intake in preschool-aged children with cerebral palsy: relationship to oropharyngeal dysphagia and functional gross motor skills. **Katherine Benfer**, Kelly Weir, Kristie Bell, Robert Ware, Peter Davies, Roslyn Boyd. 7th Biennial

Conference of the Australasian Academy of Cerebral Palsy and Developmental Medicine, March 2014.

7. Food textures habitually consumed by preschool-aged children with cerebral palsy: relationship to oropharyngeal dysphagia and gross motor functional skills. **Katherine Benfer**, Kelly Weir, Kristie Bell, Robert Ware, Peter Davies, Roslyn Boyd. 7th Biennial Conference of the Australasian Academy of Cerebral Palsy and Developmental Medicine, March 2014.
8. Early natural history of cerebral palsy: comprehensive outcomes of the Australian Cerebral Palsy child studies [invited workshop]. Roslyn N Boyd, Kristie Bell, Rachel Jordan, **Katherine Benfer**, Stina Oftedal, Andrea Coleman, Jaqueline Walker, Koa Whittingham, Kelly Weir. 7th Biennial Conference of the Australasian Academy of Cerebral Palsy and Developmental Medicine, March 2014.
9. Functional oropharyngeal impairments and their relationship to gross motor skills in young children with cerebral palsy. **Katherine Benfer**, Kelly Weir, Kristie Bell, Peter Davies, Robert Ware, Roslyn Boyd. 67th Annual Meeting of the American Academy of Cerebral Palsy and Developmental Medicine, 16-19 October 2013.
10. Food textures habitually consumed by preschool-aged children with cerebral palsy: relationship to oropharyngeal dysphagia and functional gross motor skills. **Katherine Benfer**, Kelly Weir, Kristie Bell, Peter Davies, Robert Ware, Roslyn Boyd. 67th Annual Meeting of the American Academy of Cerebral Palsy and Developmental Medicine, 16-19 October 2013.
11. Oropharyngeal dysphagia and its relationship to dietary intake and gross motor functional skills in young children with cerebral palsy. **Katherine Benfer**, Kelly Weir, Kristie Bell, Peter Davies, Robert Ware, Roslyn Boyd. Speech Pathology Australia National Conference, 24-26 June 2013.
12. Oropharyngeal dysphagia on food and fluid textures in young children with cerebral palsy: a comparison between direct clinical assessment and parent report. **Katherine Benfer**, Kelly Weir, Kristie Bell, Peter Davies, Robert Ware, Roslyn Boyd. Speech Pathology Australia National Conference, 24-26 June 2013.
13. Reported and observed clinical signs of oropharyngeal aspiration in young children with cerebral palsy. **Katherine Benfer**, Kelly Weir, Roslyn Boyd. 6th

Biennial Conference of the Australasian Academy of Cerebral Palsy and Developmental Medicine, 30 May-1 June 2012.

14. Subtypes of Oral Motor Dysfunction in feeding and its relationship with gross motor skills in young children with cerebral palsy. **Katherine Benfer**, Kelly Weir, Kristie Bell, Peter Davies, Robert Ware, Roslyn Boyd. 6th Biennial Conference of the Australasian Academy of Cerebral Palsy and Developmental Medicine, 30 May-1 June 2012.
15. Oral Motor Dysfunction on food and fluid textures, and its relationship with gross motor skills in children with cerebral palsy. **Katherine Benfer**, Kelly Weir, Roslyn Boyd. 6th Biennial Conference of the Australasian Academy of Cerebral Palsy and Developmental Medicine, 30 May-1 June 2012.
16. Subtypes of Oral Motor Dysfunction in feeding and its relationship with gross motor skills in young children with cerebral palsy. **Katherine Benfer**, Kelly Weir, Kristie Bell, Peter Davies, Robert Ware, Roslyn Boyd. 6th Biennial Conference of the Australasian Academy of Cerebral Palsy and Developmental Medicine, 30 May-1 June 2012 [poster].
17. Oral feeding ability on food and fluid textures, and their relationship with gross motor skills in young children with cerebral palsy. **Katherine Benfer**, Kelly Weir, Kristie Bell, Peter Davies, P. Robinson, Robert Ware, Roslyn Boyd. 65th Annual Meeting of the American Academy of Cerebral Palsy and Developmental Medicine, 12-15 October 2011.
18. Reported and observed clinical signs of oropharyngeal aspiration in young children with cerebral palsy. **Katherine Benfer**, Kelly Weir, Kristie Bell, Peter Davies, P. Robinson, Robert Ware, Roslyn Boyd. European Academy of Childhood Disability, 8-11 June 2011 [poster]
19. Oral feeding ability on food and fluid textures, and their relationship with gross motor skills in young children with cerebral palsy. **Katherine Benfer**, Kelly Weir, Kristie Bell, Peter Davies, Priscilla Robinson, Robert Ware, Roslyn Boyd. European Academy of Childhood Disability, 8-11 June 2011 [presented by Kelly Weir on behalf of first author]
20. Oral motor feeding skills and their relationship with gross motor skills in young children with cerebral palsy. **Katherine Benfer**, Kelly Weir, Kristie Bell, Peter Davies, Priscilla Robinson, Robert Ware, Roslyn Boyd. European Academy of Childhood Disability, 8-11 June 2011 [poster]

Invited Speaker:

1. Oropharyngeal dysphagia: Relationship to gross motor attainment in young children with cerebral palsy Paediatric Dysphagia Special Interest Group, National Telepresentation, July 2012

Awards and Scholarships:

1. Graduate School International Travel Award, 2013 ($3200)
2. American Academy of Cerebral Palsy and Developmental Medicine Annual Meeting 2014 Student Scholarship ($1050 plus conference registration)
3. American Academy of Cerebral Palsy and Developmental Medicine Annual Meeting 2013 Student Scholarship ($1000 plus conference registration)
4. Speech Pathology Australia Postgraduate Student Research Grant, 2012 ($2000)
5. American Academy of Cerebral Palsy and Developmental Medicine Annual Meeting 2011 Student Scholarship ($1000 plus conference registration)

Bell *et al. BMC Public Health* 2010, **10**:179
http://www.biomedcentral.com/1471-2458/10/179

BMC Public Health

STUDY PROTOCOL Open Access

A prospective, longitudinal study of growth, nutrition and sedentary behaviour in young children with cerebral palsy

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Abstract

Background: Cerebral palsy is the most common cause of physical disability in childhood, occurring in one in 500 children. It is caused by a static brain lesion in the neonatal period leading to a range of activity limitations. Oral motor and swallowing dysfunction, poor nutritional status and poor growth are reported frequently in young children with cerebral palsy and may impact detrimentally on physical and cognitive development, health care utilisation, participation and quality of life in later childhood. The impact of modifiable factors (dietary intake and physical activity) on growth, nutritional status, and body composition (taking into account motor severity) in this population is poorly understood. This study aims to investigate the relationship between a range of factors - linear growth, body composition, oral motor and feeding dysfunction, dietary intake, and time spent sedentary (adjusting for motor severity) - and health outcomes, health care utilisation, participation and quality of life in young children with cerebral palsy (from corrected age of 18 months to 5 years).

Design/Methods: This prospective, longitudinal, population-based study aims to recruit a total of 240 young children with cerebral palsy born in Queensland, Australia between 1st September 2006 and 31st December 2009 (80 from each birth year). Data collection will occur at three time points for each child: 17 - 25 months corrected age, 36 ± 1 months and 60 ± 1 months. Outcomes to be assessed include linear growth, body weight, body composition, dietary intake, oral motor function and feeding ability, time spent sedentary, participation, medical resource use and quality of life.

Discussion: This protocol describes a study that will provide the first longitudinal description of the relationship between functional attainment and modifiable lifestyle factors (dietary intake and habitual time spent sedentary) and their impact on the growth, body composition and nutritional status of young children with cerebral palsy across all levels of functional ability.

Background

Cerebral palsy (CP) is the most common cause of physical disability in childhood occurring in 1 in 500 children [1]. It is a group of permanent disorders of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing foetal or infant brain [2]. Damage to the structure of the brain is static and permanent; however, the consequent symptoms are variable and may change over time [2]. In addition to disordered movement or posture, children may have a range of associated disabilities, including intellectual disability, hearing and visual deficits, nutrition, feeding and swallowing problems, respiratory infections and epilepsy [1]. Cerebral palsy has substantial life long effects on daily function and quality of life (QOL) for children and their families with an estimated economic cost of over AUD $115,000 per person per annum [3].

Growth and nutritional status of children with CP

Poor growth and nutritional status are commonly reported in children with CP [4,5]. Conversely, there is evidence to suggest that certain children with CP are at risk of obesity, particularly those with marked spasticity and who are relatively inactive [6]. Poor growth is frequently considered a 'normal', untreatable side-effect of

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CP, however, the impact of poor growth on health, participation and QOL is an area that requires further investigation [7]. Most studies have concentrated on severely impaired children and are frequently flawed by a lack of valid and repeatable methods for assessing linear growth and body composition in this population [8].

The largest study to date into the growth parameters of children and adolescents with CP was based on retrospective data relating to height and weight obtained from the patient records of 24,920 children and adolescents aged 2 - 20 years. The 10th, 50th and 90th percentile curves for body weight, height and body mass index (BMI) were developed from over 141,900 measurements of weight and height [8]. This study confirmed that children with moderate to severe motor impairment are growth impaired. Trends for lower weight and height for age were apparent for the lower functioning groups and deviated further from those of the general population with increasing functional impairment. The largest differences in weight and height were seen in those with the most severe motor impairment. Interestingly, in the lowest functioning groups (groups 4 and 5), the presence of a feeding tube was associated with greater weight and height (group 5), in comparison to children in group 4 who did not have a feeding tube. The major strengths of this study were the large sample and the development of growth curves stratified by gross motor skills and mode of feeding. The most significant limitations of the study were that the methods utilised to measure height were of unknown validity and reliability in this population, non validated tools were used to determine functional severity and the sample was largely cross sectional with only a portion having repeated measures.

Importantly, the growth curves presented in this study are purely descriptive of growth within the study population and have not been related to health outcomes [8]. Any representative sample of children with CP will include a large number of undernourished subjects, as such, these population specific growth charts are not a prescription for desirable growth in this group. This study raises two key questions related to growth and nutrition for children with CP: what is desirable growth and, what is the relationship between growth, nutritional status and health related outcomes and QOL in this population?

Causes of poor growth in CP

It has been hypothesised that poor growth in children with CP may be related to nutritional factors, physical factors or factors related to the brain lesion itself. Nutritional factors include inadequate dietary intake, secondary to impaired oral motor and swallowing competence and poor nutritional status and may impact directly on growth [4,9,10]. Physical factors result in decreased mechanical stress on bones due to immobility or lack of weight bearing [11]. Bone growth studies have suggested that immobilisation decreases bone formation and longitudinal bone growth and increases bone resorption, which suppresses certain growth-stimulating hormones [11]. Factors related to the brain lesion itself may impact on growth either directly (via a negative neurotrophic effect on linear growth) or indirectly (via the endocrine system) [4,10]. Growth differences between impaired and unimpaired limbs in children with hemiplegia, support the hypothesis that non-nutritional factors play a significant role in reducing growth in children with CP [12].

Cross-sectional studies have identified links between feeding ability and measures of growth and nutritional status [13,14]. Longitudinal investigations have found that early nutritional supplementation by gastrostomy results in improved linear growth in children with severe CP if commenced early in life [10,15,16]. Swallowing difficulties have been reported in up to 99% of children with CP classified as Gross Motor Function Classification System (GMFCS) IV or V, the majority of which exhibit moderate to severe (76%) or profound (15%) dysphagia [17]. The prevalence of dysphagia in children with more mild motor impairment (GMFCS scores I-III) is unknown, as is the point at which oral motor dysfunction begins impacting on dietary intake and growth. Specific issues related to oral motor and swallowing problems in CP include poor saliva control and drooling [18]; difficulty sucking, chewing and swallowing [19-21]; and oropharyngeal aspiration [22-24], all of which impact on lifestyle. Poor saliva control has been associated with health and lifestyle impacts such as poor hygiene, reduced social acceptability of anterior drooling, reduced social interaction and self-esteem, increased daily cares [25-27] and aspiration of posterior drooling with associated pulmonary complications [28]. The impact of oral motor impairment on feeding and swallowing has been associated with reduced dietary intake leading to suboptimal nutritional status and requirement for tube feeding [13,14]. Other health issues related to oral motor and swallowing problems include pulmonary complications and pneumonia associated with oropharygneal aspiration requiring multiple hospitalisations [28-30], and lifestyle impacts on the child and family such as extended length of mealtimes [31].

In a sample of 171 children with CP, Stevenson and colleagues [4] found that children with severe gross motor impairment had significantly lower height *Z*-scores than less impaired children and that mid arm circumference and tricep skinfold thickness highly correlated with both height and weight *Z*-scores. This study suggests that growth is related to body composition and severity of CP. Stallings and colleagues [9] found that disease severity variables (oral motor function, ambulatory status, and

gastrostomy feeding) and non-disease variables (age, pubertal status, gender, and mid parental height) explained approximately 70-75% of the variability in length of 142 children with quadriplegic CP. After controlling for these, body composition (upper arm muscle area and percent body fat) explained 10-15% of the remaining variation. The magnitude of the impact of body composition on linear growth was similar to that of disease severity. Importantly, body composition had a stronger effect on the growth of younger children compared to older children. Both of these studies were cross sectional and therefore the strength of evidence is low.

The cross-sectional multi-centred study North American Growth in CP Project (NAGCPP) showed a significant relationship between functional severity and nutritional status in a group of 235 moderately to severely impaired children (GMFCS III-V) [32,33]. These children, aged 2-18 years, had lower fat-stores, shorter stature, and decreased muscle mass compared to typically developing children. In addition, these studies demonstrated an association between overall growth status and increased health care use and impaired participation [7,32]. The NAGCPP did not include an entire population based sample, few children were less than 3 years and only children with moderate to severe motor impairment (GMFCS III-V [34]) were included. In addition, lifestyle factors (dietary intake and time spent sedentary) were not assessed.

Physical activity and time spent sedentary in children with CP

Habitual physical activity is an established determinant of health and, in Australia, the cost of illness directly attributable to insufficient activity is AUD$377m per annum across the entire population [35]. In children, physical activity is required for healthy growth and development, including building strong bones and muscles, improving balance, and acquiring and developing motor skills [36]. The best available evidence indicates that people with mobility impairment are among the least physically active groups in society [37,38], and consequently children with CP may be at risk of sub-optimal growth and development secondary to physical inactivity. Unfortunately studies investigating the link between time spent sedentary and growth and development in young children with CP- particularly those who are unable to walk - have not been conducted. Studies which accurately document patterns of sedentary behaviour in this population and relate the data to health outcomes are urgently needed. Such studies require the development and evaluation of methods for assessing activity and inactivity in children who move in a range of different ways including crawling, cruising, rolling and bottom shuffling. Results will permit ascertainment of the importance of inactivity prevention and physical activity promotion strategies in the management of children with CP, as well as the identification of high need groups within the CP population.

Difficulties with the assessment of growth and nutritional status in CP

The neuromuscular complications associated with CP make accurate anthropometric and body composition measurements difficult and sometimes impossible in this population. Our group, and others, have overcome this issue by using segmental limb measures which provide reliable, valid and clinically useful alternatives to measuring height in children with CP [39-41]. For evaluation of body composition, the use of deuterium-oxide is considered a "gold-standard-technique" due to its reliability, accuracy and the limited assumptions required with its use compared to other more commonly used and widely available measures such as skinfold thicknesses; however, its limited availability, cost and time required for analysis result in a technique that is generally prohibitive for routine clinical use. When used in combination with published hydration constants [42,43], deuterium-oxide can be used to determine fat free mass and hence fat mass in children, using the two component model of body composition. It is a safe, non-radioactive, naturally occurring, isotope that has been used to measure total body water in a wide range of groups including pregnant women, infants and the elderly [44,45].

Current investigations into the growth, oral motor and feeding difficulties and nutritional status of children with CP have focused on cross sectional data at one time point or diverse samples across a broad age range. They concentrate on only the most severely impaired children without use of validated measures of height, body composition, gross motor function and health status. Measures of feeding ability and oral motor dysfunction have been most commonly derived from parent questionnaires rather than the use of validated clinical tools. There have been no longitudinal investigations into the impact of lifestyle factors (dietary intake and time spent sedentary) on growth, body composition, nutritional status and their impact on health outcomes in children with CP. The paucity of such information reduces capacity to develop and implement effective management strategies for this population.

Aims and hypotheses

This study will investigate the influence of growth, body composition, dietary intake, oral motor and swallowing function and time spent sedentary (adjusted for motor severity) on health outcomes, participation and QOL in a prospective population based study of young children with CP (from corrected age (ca) of 18 months to 5 years). The hypothesized interaction between these factors is

represented graphically in the conceptual model (see figure 1).

This broad aim will be addressed by the following four hypotheses (H):

H1

Growth status, nutritional status and growth velocity, from 18 months of age will be related to the level of gross motor functional attainment (GMFCS) at 5 years of age.

H2

Body composition (fat free mass and fat mass) will be related to the level of gross motor functional attainment at 5 years of age.

H3a

For a given GMFCS level, dietary intake, oral motor/swallowing function and time spent sedentary at 3 and 5 years of age will be significantly related to growth velocity and body composition.

H3b

The relationship between dietary intake, oral motor/swallowing function & time spent sedentary at 18 months will predict growth status, nutritional status and body composition at 5 years of age.

H4

Controlling for functional severity, children with slower growth, suboptimal body composition (fat free mass and fat mass), lower levels of oral motor/swallowing function and greater time spent sedentary will have:

(i) higher health care utilisation and direct medical costs at 3 and 5 years.

(ii) lower levels of participation in school, leisure and community at 3 and 5 years.

(iii) poorer QOL at 5 years.

Study significance

This study will be the first longitudinal, population based study to enable more accurate prediction of the early natural history of nutritional and growth problems in young children with CP linked to dietary intake, time spent sedentary, health outcomes and resource utilization. Specifically this project will:

- Determine the nature and timing of nutritional, feeding and growth abnormalities.
- Enable better prediction of the likelihood and impact of sub-optimal dietary intake from an earlier age.
- Enable planning of nutritional treatment options at optimal times.
- Develop and validate methods for measuring sedentary behaviour in young children with CP, including those who do not walk as their primary means of locomotion.
- Highlight the relative contribution of poor dietary intake, oral motor and feeding difficulties and sedentary behaviour on growth and body composition taking into account severity of disability.
- Quantify the impact of dietary intake and time spent sedentary on medical resource use to inform service provision planning.
- Define the relationship between habitual time spent sedentary and functional abilities to predict eventual functional attainment.
- Define the relationship between oromotor/swallow dysfunction and gross motor attainment.
- Quantify the impact of poor nutrition and high amounts of time spent sedentary on participation in society and QOL.

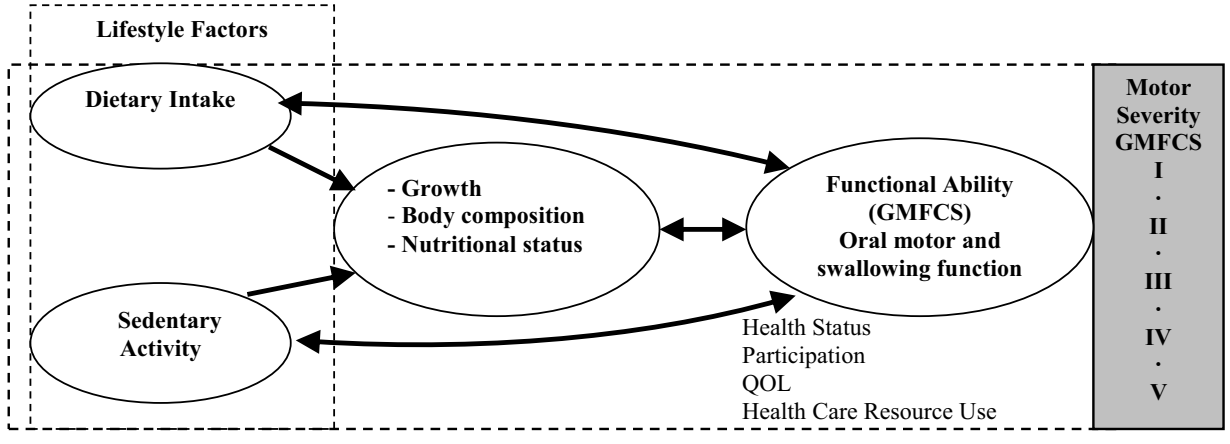

Figure 1 Conceptual model illustrating the hypothesized interactions of the principle factors to be investigated in the prospective, longitudinal study of growth, nutrition and sedentary activity in young children with cerebral palsy.

• Allow interpretation of data derived from clinical methods for the assessment of body size and composition (eg body mass index (BMI) and skin-fold thickness).

Methods/Design

This prospective, population based longitudinal study aims to recruit a total of 240 young children with CP born in Queensland, Australia, between 1st September 2006 and 31st December 2009. It is being conducted in conjunction with another study: Queensland CP Child Study of Motor Function and Brain Development (NHMRC 465128). Ethics approvals have been received from the University of Queensland Medical Research Ethics Committee (2008002260), the Children's Health Services District Ethics Committee (HREC08/QRCH/112/AM01), the CP League of Queensland (CPLQ 2008/2010 1029), Gold Coast Health Service District Human Research Ethics Committee (HREC/09/QGC/88), and the Townsville Health Service District Human Research Ethics Committee (HREC/09/QTHS/96). Further ethics approvals are being sought from additional paediatric and regional centres throughout Queensland.

Selection criteria

Inclusion criteria

All Queensland born children diagnosed with CP, born between 1st September 2006 and 31st December, 2009. We define CP as a group of permanent disorders of movement and posture that are attributed to non-progressive disturbances that occurred in the developing foetal or infant brain [2]. The characteristic signs are spasticity, movement disorders, muscle weakness, ataxia and rigidity [46].

Exclusion criteria

Children with a progressive or neurodegenerative lesion will be excluded from the study.

Recruitment

Recruitment for this study commenced in April 2009 and state-wide recruitment has been established in collaboration with the Queensland CP Register, the Queensland CP League, the Queensland Children's Health Services District, the Queensland CP Health Service, and other regional hospitals and health service districts throughout Queensland. Community awareness has been generated through paediatricians, general practitioners, allied health professionals, child health nurses, and neonatal follow-up clinics. These groups have been encouraged to refer children with motor delay (not sitting at 10 months, not standing at 12 months or walking at 24 months) for confirmation of a diagnosis of CP. Specialist clinics have been established within the Children's Health Services District where suitability for the study can be confirmed.

Study entry

Eligible children will enter the study from 18 months corrected age. They will be assessed for diagnostic criteria, co-morbidities and for differential neurological assessment by a Paediatric Rehabilitation Specialist and/or a Paediatric Neurologist. All measures will be performed on three occasions at 17 to 25 months (according to study entry); 36 ± 1 months and 60 ± 1 months corrected age (see Figure 2 flow chart for details). Children diagnosed after 25 months of age may enter the study at either 30 ± 1 months or 36 ± 1 months. To ensure collection of data at three time periods, these children will have their second assessment conducted at 48 ± 1 months. Written informed consent will be obtained from the parents or legal guardians prior to the commencement of data collection.

Feasibility

Children who are detected after 18 months of age will be entered into the study at the time of diagnosis, will receive assessment at entry and be followed up until outcome at 5 years. According to the Queensland CP Register there are 80-120 new children with CP born in Queensland each year. We propose recruitment of at least 80 children each year (total 240 children from 360 potential children). High ascertainment is expected for children with moderate to marked motor delay (GMFCS III to IV) and this has been the case for children born preterm and children referred to the Queensland CP Health Service. Children born at term with mild motor delay (GMFCS I and II) and predominant lower limb involvement (diplegia) are typically identified through the Qld CP Health Service and CP Orthopaedic services at the Royal Children's and Mater Children's Hospitals.

Recruitment and data collection for this study is being conducted in conjunction with the Queensland CP Child Study of Motor Function and Brain Development (NHMRC 465128). This is a population based prospective cohort study (n = 240) which aims to determine the pathway(s) to motor outcome (gross and fine motor) from diagnosis at 18 months to outcome at 5 years in relation to the nature of the brain lesion (using structural MRI). Children enter the study at 18 months of age with assessments conducted every six months until 3 years of age then again at 4 years and final outcomes are assessed at 5 years.

Measurements and procedures

Gross motor function

Gross Motor Function will be determined using the Gross Motor Function Measure (GMFM 66). The GMFM 66 has been shown to be valid and reliable and has been Rasch analysed to enable improved scaling [47]. Gross motor function assessment will be conducted by two

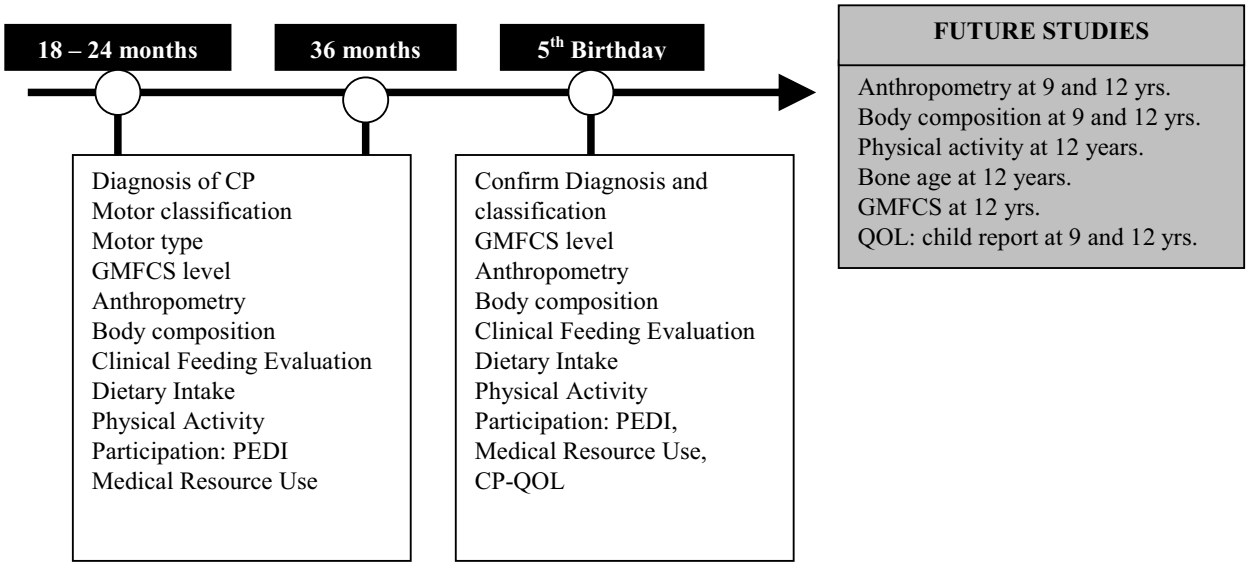

Figure 2 Flow chart of study timeline & measures from baseline (left), to completion (right). GMFCS = the Gross Motor Function classification System; FMS = Functional Mobility Scale: TBW = Total Body Water; PEDI = The Paediatric Evaluation of Disability Inventory; CAPE = the Children's Assessment of Participation and Enjoyment; CP-QOL-child = the Cerebral Palsy Quality of Life measure (child version); Dx = Confirmed diagnosis.

experienced paediatric physiotherapists whom have criterion rating with the study developers (Boyd). All GMFM assessments will be video taped to enable scoring of the accelerometry data for validation of the Actigraph for the identification of time spent sedentary.

Motor type

Type of CP (eg, spastic, dystonic, or hypotonic) and motor distribution (unilateral, bilateral, number of limbs involved) will be determined by two independent physiotherapists at each assessment according to Sanger [46] and the internationally accepted classification system on the European CP Register [48]. The classification of motor type will be recorded for both physiotherapists, independently, at each assessment and common agreement will be assessed for rating motor type at a young age.

Functional severity

Functional severity will be determined using the internationally accepted Gross Motor Function Classification System (GMFCS) [34] by two independent physiotherapists trained in performing the gross motor function assessment. Children will be classified as being in one of five functional categories for the age bands under two years, two to four years and four to six years. The GMFCS has established validity and reliability for use in young children with CP [34,49]. Inter rater reliability for the current study will be determined.

Anthropometry

Weight will be measured to the nearest 100 grams using chair scales (Seca Ltd, Germany). Height or length will be measured to the last completed millimetre with a portable stadiometer/length measuring board (Shorr Productions, LLC, Maryland, USA). Knee height and upper-arm length will be measured with an anthropometer (Holtain Ltd, Dyfed, UK). Intraobserver reliability has technical errors of 0.23 cm for upper-arm length and 0.16 cm for knee height with coefficients of variation of 1.22% and 0.56% respectively [39]. Height estimates will be predicted from knee height and upper arm length using published validated equations [39]. Body mass index will be calculated as weight (kg) divided by height (m) squared. Head circumference and mid arm circumference will be measured to the last completed millimetre using a steel flexible measuring tape.

Duplicate measurements of tricep skin-fold thickness and subscapular skin-fold thickness will be measured using callipers (Holtain Ltd, Dyfed, UK) by trained investigators. By convention, all measurements will be conducted on the left side of the body. This protocol is modelled on the convention used for development of the National Centre for Health Statistics charts [50]. Data from skin-fold thicknesses have been found to be useful when assessing the nutritional status of children with CP [14]. Reliabilities have technical errors for intraobserver and interobserver measures of tricep skin-fold thickness of 0.60 mm and 0.55 mm with coefficients of variation of 5.93% and 6.98% respectively [7].

Anthropometric and body composition data will be converted to *Z*-scores using age and gender specific reference data for the general population [51,52]. Between-group comparisons will be conducted across GMFCS levels (I-V).

Body composition

Total body water (TBW) will be measured non-invasively, using the deuterium-dilution technique [53]. Children will be given a dose of deuterium in the form of water either orally or via feeding tube. In the absence of a feeding tube in children with feeding difficulties, children will be assessed to determine the most suitable technique to enable the consumption of the isotope with minimal risk of spillage. Any spillage that may occur will be collected in an absorbent cloth which will be weighed before and after dosing to accurately determine how much fluid has been lost [54]. A single baseline urine sample will be collected prior to administration of the dose to determine natural baseline enrichments of the isotopes and a second urine sample will be collected at approximately five hours after dosing. Measurement of the isotopic enrichment of a sample of body fluids at this time enables calculation of the body water pool using standard equations [55]. Collection of urine samples from children with poor or no bladder control will involve the inclusion of an absorbent liner in their nappy from which urine will be extracted for analysis [54]. Analyses of the urine samples will be performed using an isotope ratio mass spectrometer. Similar procedures have been used by our group and others in infants, children following severe traumatic brain injury and children with mild and severe CP [53,56-58]. The accuracy of TBW measured using the deuterium dilution technique is excellent at approximately 1% [59], and 1 - 2% for repeated measurements [60,61]. Fat free mass will be determined through division of TBW by age and gender specific hydration factors [42].

Bioelectrical impedance analysis

Impedance (Ohm) will be measured using a Body Stat 1500MDD (Isle of Mann, UK) at 800μA and a fixed frequency of 50 KHz. Children will be required to lie in a supine position with arms and legs slightly abducted from the trunk. The electrical current will be applied through two non-polarizing surface electrodes placed at the dorsal surfaces of the hand and foot over the distal aspect of the second and third metacarpals and metatarsals. The voltage drop will be measured by two further electrodes placed at the right pisiform prominence of the wrist and between the lateral and medial malleoli of the ankle. The proximal and distal electrodes will be a minimum of 5 cm apart. All measurements will be taken twice, with a third measurement taken if the difference is greater than 5 Ohm. The mean of the two closest values will be used for analysis. Total body water will be estimated from measurement of impedance and height or length using previously published equations [62-64]. The relationship between height^2/impedance and TBW measured using deuterium dilution will be examined using regression analysis. An equation for the estimation of TBW from measures of height or length and impedance, specific for young children with CP, will be developed [53]. Reliability of measurements of impedance in this population will be determined.

Habitual time spent sedentary

The time that children spend sedentary in their own free-living environment will be measured using the ActiGraph GT3M accelerometer (Shalimar, FL). The GT3M is a small (3.8 × 3.7 × 1.8 cm), lightweight (27 g) triaxial accelerometer that detects accelerations of a magnitude and frequency that correspond with human movement, filtering out other forms of motion (e.g. vibration). Raw acceleration data is recorded in real time as counts per minute. Output from the device can be used to indicate when the wearer was active, as well as when they were sedentary.

Accelerometry is the most appropriate method for measurement of sedentary behaviour in this study. Self-report is inappropriate in this age-group as our pilot data demonstrate that parental report correlates poorly with criterion measures in the target population [65]. Pedometers measure only steps and are therefore inappropriate for use with children who do not walk, and doubly labelled water, while considered the gold standard for the measurement of physical activity, is prohibitively expensive and will not provide data on patterns of activity. Additionally, the ActiGraph has demonstrated cross-validity with criterion measures of activity in populations, age groups and activities relevant to the current study including: hip worn ActiGraphs for walking people with brain injury (r = 0.74) [66]; for measuring free play in young children (r = 0.72) [67]; and wrist worn ActiGraphs for measuring wheelchair activity in people with disabilities (r = 0.66) [68].

Time spent sedentary vs time spent active While both time spent active and sedentary behaviour have established links with child health outcomes [36], our study will focus on measurement of sedentary behaviour. Time spent active will not be used as an outcome measure as young children with CP move in a variety of ways including walking, running, crawling, creeping, rolling, and bottom-shuffling. In combination with disordered movement kinematics and kinetics, these diverse modes of movement make the relationship between counts per minute and activity intensity for children with CP unpredictable. As a consequence, identification of time spent in moderate to vigorous physical activity (the intensity recommended for normal growth and development [36]) is impossible to derive from accelerometer output in this population. In contrast to accelerometer-based measurement of activity, we can be confident that, providing a child is wearing the monitor, if counts per minute are zero, the child is sedentary.

Identification of cut points for sedentary behaviour

There is a methodological challenge in choosing to measure sedentary behaviour: when counts per minute are

greater than zero it does not necessarily follow that the child is active (e.g., very low but non-zero counts per minute will be registered with regular weight-shift that occurs with prolonged sitting). Therefore, in order to validly determine when a child has been sedentary, a criterion validity study will be conducted to determine cut-points for differentiating between sedentary behaviour and non-sedentary behaviour. The method used will be based on that described by Welk et al [69]

Participants in the criterion validation study for the Actigraph will be 100 children with CP participating in the Queensland CP Child Study of Motor Function and Brain Development, with a minimum of two children in each of the 15 possible combinations of age (17 to 25 months, 36 ± 1 months and 60 ± 1 months) and GMFCS level (I-V) in our sample. As part of their evaluation, children in the Queensland CP Child: Brain and Motor Development Study complete the Gross Motor Function Measure 66 [47], a standardised motor assessment battery which takes between 40-60 min to complete and requires the children to complete a range of motor tasks (e.g., sitting, standing, rolling, crawling etc). During these assessments, children will wear an Actigraph GT3M and will be video taped. The video of the assessment will subsequently be coded using BEST direct observation software to provide a real-time criterion measure of when the child was active and when they were sedentary. Active behaviour is defined as either positional change where the centre of gravity is moved (e.g., sit to stand, stand to sit, bending down) or translocation of any description (e.g., walking, crawling, rolling). Sedentary behaviour is defined as the child being stationary with or without limb or head movement. To derive unique sedentary cut-points (count per minute) which maximize sensitivity and specificity in each of the 15 cells, a receiver operator characteristic curve analysis will be conducted. For this analysis, counts per minute will serve as the independent variable, with a (1, 0) indicator variable corresponding to 1 = sedentary (as determined from direct observation) versus 0 = non-sedentary activity (again, as determined from direct observation) serving as the dependent variable.

Measurement of habitual time spent sedentary To measure free-living sedentary behaviour at each of the three planned data collection points, ActiGraphs will be set for 15 second epochs and worn at the centre of the child's back [70], for a period of 3 days, the minimum required for a valid estimate of habitual activity in children [70]. Children will be required to wear the Actigraph during waking hours only and parents will be given instructions for wear and logging wear-time. After 3 days, the ActiGraph will be returned by courier for data extraction and analysis. Following return, output will be analysed for periods of non-wear and the data converted to mean counts per minute for the monitoring period. Analysis will be performed according to functional severity (GMFCS) with mean counts per minute used to stratify participants into high, medium and low levels of sedentary behaviour.

Dietary intake

Usual dietary intake will be determined using a three day weighed food record [71]. Parents will be instructed to weigh all food and fluids offered to the child before and after consumption. Parents will also be instructed to record information regarding the amount of food and fluids lost due to spillage as well as the time taken (in minutes) for the child to consume each meal, snack or drink. Food records will be reviewed by the Research Dietician with the caregiver present to clarify any ambiguous information. Food records will be analysed using the Foodworks™ dietary analysis software program (Xyris Software (Australia) Pty Ltd). Mean energy intake will be expressed as a percentage of age and gender specific recommendations [72].

Feeding ability

Oral motor and swallowing function will be assessed using a number of measures obtained from a parent completed feeding questionnaire, direct observation during a clinical feeding evaluation of a regular meal and from ratings derived from the video taped clinical feeding evaluation. Saliva control and drooling measures were derived from parent report and clinician's rating during the clinical feeding evaluation using a five point scale for severity and four point scale for frequency described by Thomas-Stonell and Greenberg [73]. A subset of clinical signs suggestive of aspiration will be noted from parent report in the feeding questionnaire and during the clinical feeding evaluation [74]. Objective measurement of oral motor function during feeding will be rated from the videotaped clinical feeding evaluation using the Schedule for Oral Motor Assessment (SOMA). The SOMA was normalised on 127 young infants aged 8-24 months with 10% of the population having CP. It has a positive predictive validity of 90% and sensitivity greater than 85% to detect clinically significant oral-motor dysfunction in infants and young children. This assessment has also been used to evaluate children of older ages. The SOMA has excellent levels of inter-rater reliability (kappa > 0.75) and intra-rater reliability (85%) [75-77]. Oral motor and swallowing function will also be formally rated using the Feeding and Swallowing Competency Subtest (Part 2) of the Dysphagia Disorders Survey (DDS) - Pediatric [17,78]. The DDS was developed as a screening tool to assess feeding and swallowing function in children and adults with developmental disability [3-78 years; mean 31.71 years] with 5% of the population aged 3-17 years (n = 31). It has more recently been used in a group of 166 children (2 years 1 month - 19 years 1 month; mean 9 years 4

months) with moderate to severe CP and intellectual disability [17,78]. Test validity and inter-item reliability were determined from a sample of 626 people with developmental disability. Inter-rater reliability was undertaken on a sample of 21 participants by 6 speech pathologists and achieved excellent reliability of 97% [17,78]. Inter-rater reliability of direct ratings for the SOMA and DDS will be compared for 10% of the participants in our study.

Participation

Participation will be determined using parent-report on the domains of self-care, mobility and social functioning using the scaled scores (rasch analysed) of the Pediatric Evaluation of Disability Inventory (PEDI) [79]. The PEDI is a generic standardised instrument of functional performance in children with disabilities that has been found to be both valid and reliable. It has been standardised on a sample of 412 able bodied American children between the ages of 0.5 and 7.5 years [79]. There are three independent domains of the PEDI (participation in self-care, mobility and social function) that are rated by parent report as capable (score = 1) or incapable to perform (score = 0). The PEDI has been found to be a valid and reliable assessment of functional performance in children with disabilities [79].

The mobility and self-care domain of the PEDI will be completed by the caregiver to assess the child's participation in activities of daily living. On the first occasion the PEDI will be administered as an interview (15-20 mins). On subsequent occasions it will be provided as a questionnaire mailed to the family for completion prior to the study visit, and will be checked by the researcher at the study visit. The PEDI raw aggregate scores can be converted into normative standard scores and scaled scores using conversion tables provided in the manual [79]. Scaled scores provide an indication of the child's performance along a continuum of item difficulty or complexity in a particular domain. The range of possible scores (0-100) represents increasing levels of function. In the present study, all raw scores will be converted to scaled scores (Rasch analysed) to compare the entire group (age range 18 months to five years) of all children across the self-care domain for capability, without the difficulties of 'ceiling and floor' effects due to age limitation in the normative standard scores.

Quality of life

Parent perception of QOL will be assessed using the condition specific tool CP QOL-child (CP QOL-Child) from 4 years of age [80]. The CP QOL-Child assesses aspects of life that parents and children have identified as important including physical wellbeing, social wellbeing, emotional wellbeing, school, access to services, and acceptance by others. The psychometric properties of the CP QOL - Child are excellent with Cronbach's Alpha range from 0.74-0.92 for parent-proxy report [80]. Test re-test is adequate, where ICC 0.76-0.89 and it is moderately correlated with generic QOL and health (r = 0.30-0.51) [80]

Resource use and the direct costs of treatment

In order to determine the relationship between motor prognosis and resource use, medical and allied health resource use and the direct costs of treatment will be monitored and compared to outcomes with adjustment for confounders such as disease severity using cost and consequences analysis [81].

Sample size calculations

240 children will be studied with three measurements planned for each participant between 18 months and 5 years of age. For hypothesis 1, a sample size of 45 per group (GMFCS I-V, totalling 225 patients) will have 80% power and 5% significance of detecting a between group difference in height of 6 cm (assuming a standard deviation of change of 10 cm) between 18 months and 5 years of age, between functional groups and non-CP infants [52]. To allow for attrition we will enrol 240 infants in total.

Statistical considerations

Primary analysis will use the intention to treat principle, using the Last Observation Carried Forward principle for participants who withdraw before the end of the study period. Differences between participants who complete and withdraw will be assessed using t-tests for continuous variables, after transformations of non-normally distributed variables, and Fisher's Exact Test for categorical variables. Baseline characteristics of the GMFCS groups will be compared similarly. Details for the statistical models that will be used to analyse data to address each hypothesis are as described below.

H1

Outcome is attainment of GMFCS, a 5-level categorical variable at 5 yrs. We will consider the explanatory variables of growth and nutritional status in separate models. Individual Z-scores for height or predicted height from knee height or upper arm length will be determined at 18, 36 and 5 yrs and modelled using mixed-effects models. These models are used as they incorporate both fixed and random variables in the analysis. We will model using a random-intercept and slope for each participant. We will test potential covariates (eg sex) and include them as fixed effects if appropriate.

H2

Outcome is attainment of GMFCS at 5 years. Explanatory variables are fat free mass and fat mass at 18, 36 and 5 years. We will investigate the association between explanatory and outcome variables using separate mixed-effects models.

H3a

Outcome variables are growth status and body composition. We will investigate the association with explanatory variables of dietary intake and habitual physical activity at 3 years and 5 years. We will use mixed-effects models with random intercept and slope for each participation, with GMFCS as a fixed effect and with appropriate interaction terms.

H3b

Outcome variables are growth status, nutritional status and body composition at 5 years of age. Explanatory variables are dietary intake and time spent sedentary at 18 months of age. We will investigate the ability of the explanatory variables to predict the outcome variables using mixed-effects models.

H4

Outcomes are health care utilization and direct medical costs, participation (PEDI) at 3 and 5 years and QOL at 5 yrs. Explanatory variables are growth, body composition and time spent sedentary at 18 months and 36 months. We will investigate the association between explanatory and outcome variables using mixed-effects models with a random intercept and slope for each participant, and functional severity at 18 months included as a fixed effect.

Competing interests

The authors declare that they have no competing interests.

Authors' contributions

PSWD, RNB, KLB, SMT, KAW and RDS contributed to the study design, study protocol and grant writing. KLB modified the grant for publication with input from all coauthors. All authors read and approved the final manuscript.

Acknowledgements

This study has been funded by the Australian National Health and Medical Research Council Project Grant (569605). RNB is funded by a National Health and Medical Research Council Career Development Grant (473480), the Royal Children's Hospital Foundation and a Smart State Fellowship through the Queensland Department of Innovation. We wish to thank our Associate Investigators Dr Robert Ware for statistical advice and input, Dr Stewart Trost and Mr Michael de Lacy, as well as Dr Lynne McKinlay, Dr Kate Sinclair and Ms Megan Kentish.

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Received: 15 March 2010 Accepted: 6 April 2010
Published: 6 April 2010

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Pre-publication history
The pre-publication history for this paper can be accessed here:
http://www.biomedcentral.com/1471-2458/10/179/prepub

doi: 10.1186/1471-2458-10-179
Cite this article as: Bell *et al.*, A prospective, longitudinal study of growth, nutrition and sedentary behaviour in young children with cerebral palsy *BMC Public Health* 2010, **10**:179

Boyd et al. BMC Neurology 2013, 13:57
http://www.biomedcentral.com/1471-2377/13/57

BMC Neurology

STUDY PROTOCOL Open Access

Australian Cerebral Palsy Child Study: protocol of a prospective population based study of motor and brain development of preschool aged children with cerebral palsy

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Abstract

Background: Cerebral palsy (CP) results from a static brain lesion during pregnancy or early life and remains the most common cause of physical disability in children (1 in 500). While the brain lesion is static, the physical manifestations and medical issues may progress resulting in altered motor patterns. To date, there are no prospective longitudinal studies of CP that follow a birth cohort to track early gross and fine motor development and use Magnetic Resonance Imaging (MRI) to determine the anatomical pattern and likely timing of the brain lesion. Existing studies do not consider treatment costs and outcomes. This study aims to determine the pathway(s) to motor outcome from diagnosis at 18 months corrected age (c.a.) to outcome at 5 years in relation to the nature of the brain lesion (using structural MRI).

Methods: This prospective cohort study aims to recruit a total of 240 children diagnosed with CP born in Victoria (birth years 2004 and 2005) and Queensland (birth years 2006–2009). Children can enter the study at any time between 18 months to 5 years of age and will be assessed at 18, 24, 30, 36, 48 and 60 months c.a. Outcomes include gross motor function (GMFM-66 & GMFM-88), Gross Motor Function Classification System (GMFCS); musculoskeletal development (hip displacement, spasticity, muscle contracture), upper limb function (Manual Ability Classification System), communication difficulties using Communication and Symbolic Behaviour Scales-Developmental Profile (CSBS-DP), participation using the Paediatric Evaluation of Disability Inventory (PEDI), parent reported quality of life and classification of medical and allied health resource use and determination of the aetiology of CP using clinical evaluation combined with MRI. The relationship between the pathways to motor outcome and the nature of the brain lesion will be analysed using multiple methods including non-linear modelling, multilevel mixed-effects models and generalised estimating equations.

Discussion: This protocol describes a large population-based study of early motor development and brain structure in a representative sample of preschool aged children with CP, using direct clinical assessment. The results of this study will be published in peer reviewed journals and presented at relevant international conferences.

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Trial registration: Australia and New Zealand Clinical Trials Register (ACTRN1261200169820)

Keywords: Cerebral palsy, Protocol, Longitudinal cohort, Motor development, Brain structure and function, Communication, Hip displacement, Preschool age, Gross motor function

Background

Cerebral Palsy (CP) is a disorder of movement and posture secondary to an insult to the developing brain [1]. The insult is static and permanent and may be the consequence of different factors, including both genetic and environmental causes. Although the insult is static, the consequent symptoms are variable and may change over time [2]. Children may have a range of associated disabilities, including intellectual disability, hearing and visual deficits, nutritional and feeding problems, respiratory infections and epilepsy [3,4]. Secondary musculoskeletal disorders involving muscle, tendons, bones and joints are common as a result of spasticity, muscle weakness and immobility. CP has substantial lifelong effects on daily function, societal participation and quality of life (QOL) for children and their families.

Cerebral Palsy registers have provided us with some understanding of the aetiologies of CP and specific outcome studies [3]. Few studies have documented broad clinical outcomes for an entire cohort of children with CP prospectively. In addition, none of the existing cohort studies have utilised their large patient groups to better understand the aetiologies of CP, the relationship between abnormalities on brain MRI and outcomes such as motor disability [5] musculoskeletal deformity and related development (communication, oromotor, fine motor skills). A better understanding of the aetiology of CP, the timing of the insult during brain development and the anatomical pattern of injury or malformation is required in order separate CP into different prognostic or treatment groups and to determine the pathway to motor outcome.

Previous studies [5-8] have reported the relative proportions of GMFCS levels (GMFCS I: 27.9-40.7%, GMFCS II: 12.2%-18.6%, GMFCS III: 13.8%-18.6%, GMFCS IV: 11.4%-20.9%, GMFCS V: 15.6%-20.5%), motor types (spastic: 78.2-86.4%, dyskinetic: 1.5%-6.1%, mixed: 6.5%-9.1%, ataxia: 2.5%-2.8%, hypotonia: 2.8%-4.1%), and motor topography (hemiplegia: 15.3%-40.0%, diplegia: 28.0%-46.4%, quadriplegia: 13.6%-50.8%) within various CP cohorts [6,9,10]. A recent systematic review investigating the rates of co-occurring impairments, diseases and functional limitations in CP concluded that for children diagnosed at 5 years of age: 3 in 4 were in pain; 1 in 2 had an intellectual disability; 1 in 3 could not walk; 1 in 3 had hip displacement; 1 in 4 could not talk; 1 in 4 had epilepsy; 1 in 4 had a behaviour disorder; 1 in 4 had bladder control problems; 1 in 5 had a sleep disorder; 1 in 5 dribbled; 1 in 10 were blind; 1 in 15 were tube fed; and 1 in 25 were deaf [4]. Launched in 2007, the Australian Cerebral Palsy Register [3] combines data from several notable state-wide registries (including Queensland, Victoria, Western Australia and New South Wales), and is one of the largest CP registers in the world with over 3,000 children registered in the 1993–2003 birth cohort.

Hip displacement is the second most common musculoskeletal problem in children with CP [11-14]. In the most severely impaired, non-ambulatory children, the incidence may be as high as 80% [11,15]. While children with CP are born with enlocated hips, progression to hip displacement is demonstrated in some children with CP from a very early age [13,14,16]. Hip surveillance programs and appropriately-timed interventions improve outcomes at skeletal maturity [14,15]. Although the final outcome of early intervention at skeletal maturity is not clear [17,18], early risk assessment might enable earlier referral for those children who may benefit from preventative intervention [19]. As clinical assessment of hip range of motion is a poor predictor of risk, several radiological and clinical measures are used to diagnose and monitor hip subluxation [13,16,17,19]. While functional disability, pain [20] and impaired ambulatory weight-bearing [12,16,18,19] are associated with risk of hip displacement and need for surgical intervention, the evidence regarding radiological characteristics is less clear [21,22]. There is a need for early prospective evaluation of radiological development in a population of very young children with CP across the spectrum of function severity in order to aid prediction of hip development.

There have been several large studies that have evaluated prospective motor development in children with CP. The Ontario Motor Study (OMGS) collated over 2,632 GMFM assessments on 657 children with an average of four observations per child [9]. The principal outcome of the study was the development of two internationally accepted valid and reliable tools for measuring motor function (the Gross Motor Function Measure, GMFM) [9,23] and for classifying functional status into five groups (Gross Motor Function Classification System, GMFCS) [24,25]. From these data, Growth Motor curves for children with CP were developed [9]. These curves are valid and reliable for children aged two years and over and allow for tracking and predicting motor

outcomes for children by GMFCS classification [25]. Two potential limitations of the Ontario Motor Study were that it included only minimal data on children less than 3 years of age and it was a not an entire population based sample [9].

In the European Cerebral Palsy study [6], with a representative cohort of children with CP from eight European countries, children are classified according to brain injury diagnosed using MRI. This group used a classification system based on the presumed timing and nature of the insult that resulted in CP and included both genetic and non-genetic aetiologies such as genetic cortical malformations (e.g. lissencephaly) and hypoxic ischaemic injury [6,10]. Again this cohort is representative rather than entire population based and these investigators from Surveillance of Cerebral Palsy in Europe (SCPE) have guided our classifications of motor type and of the brain injury on MRI [26-28].

Pathogenic events impacting on the brain cause different patterns of structural abnormality in CP [29]. These pathogenic events may be environmental or genetic. Their consequences will depend not only on the nature of the event, but also the timing of the event during the different stages of brain development (Figure 1). The 1st and 2nd trimesters are the most critical times for cortical development and are characterized by the sequential yet overlapping steps of proliferation, migration and organization of neuronal cells and their connections. Brain pathology secondary to events during these stages of brain development is usually characterised by significant malformations. During the 3rd trimester, growth and differentiation events are predominant and persist into postnatal life. Disturbances of brain development during this period cause lesions, often of a different pattern to those resulting from earlier insults or developmental disorders. During the early 3rd trimester, the periventricular white matter is especially affected; whereas towards the end of the 3rd trimester grey matter, either cortical or deep grey matter, appears to be more vulnerable. Understanding the aetiologies of CP in the living patient has advanced significantly since the increased use of MRI in the evaluation of children with congenital or early-onset neurological deficits. Using MRI, a number of studies have shown that the most common causes of CP are structural brain lesions [27,30-33], especially prematurity-related injuries, and malformations of brain development [34-36]. Guidelines by the American Academy of Neurology strongly recommend that all children with a suspected diagnosis of CP undergo neuroimaging, with MRI preferable to CT [37]. Determination of brain structural abnormality will provide a final diagnosis that is more than a label of 'cerebral palsy'[38].

It is necessary to attempt to determine the underlying aetiology/pathogenesis to confirm the suspicion of a static lesion, exclude a treatable disorder and diagnose a malformation, which may have significant genetic counselling implications for the family. In addition, these patterns of brain maldevelopments or lesions offer excellent models to study the normal mechanisms of organisation and reorganisation in the developing brain [30,31,39]. Despite these advances, limited studies exist correlating the specific MR imaging appearance and outcome measures such as motor function [27]. Such data may prove invaluable in providing accurate prognostic counselling at the time of diagnosis, as well as potentially guiding the most appropriate treatments tailored to each individual's pattern of CP and type of lesion on imaging.

A recent systematic review investigated the relationship between brain structure on MRI and motor

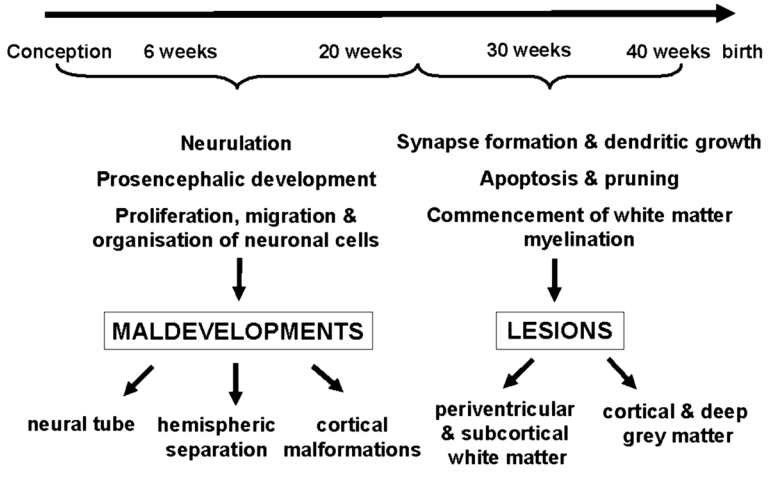


Figure 1 Major events in human brain development. Pathogenic events (both genetic and non-genetic) affect the developing brain to cause malformations or lesions, the patterns of which will depend on the stage of brain development during which the event occurs.

outcomes in children with CP [40]. A total of 37 studies comprising over 2300 subjects met inclusion criteria, and these studies were analysed in terms of population characteristics, MRI data, motor outcome data, and where possible, the relationship between MRI data and motor outcomes. The importance of MRI lesion description has been previously outlined, due to the presumed relationships between lesion topography and motor type, and between lesion extent and functional severity [27]. Indeed, Yokochi et al. [29] and Holmstrom et al. [41] reported that in subjects with motor subtypes of athetosis or hemiplegia respectively, motor disabilities were more severe when lesions involved both grey and white matter on MRI as opposed to grey or white matter involvement alone. Similarly, Holmefur et al. [42] reported that in subjects with spastic hemiplegia, those with more severe white matter reduction on MRI had a significantly lower development in hand function. A focus of current research is the prevention of CP, which requires clinical outcomes to be correlated with the presumed timing and aetiology of lesions in the developing brain [43]. Pathological insults during brain development cause abnormalities or lesions which may be detected by brain MRI, and the observable patterns of these lesions depend on the stage of brain development [39]. Using this principle, a qualitative classification system has emerged whereby lesions can be identified as brain maldevelopments, periventricular white matter lesions, grey matter lesions, other miscellaneous lesions, or normal MRI [27]. All studies included in the review reported enough MRI data for subjects to be classified into these broad lesion groups, and differences in motor subtypes and functional disabilities were identified between groups [40]. Despite this, it was found that many studies did not utilise valid and reliable classifications and measures of motor abilities (e.g. GMFCS, GMFM, and MACS), and heterogeneous measures were employed which generally precluded pooled analysis. All included studies also used a qualitative system of lesion description or classification [27], and as such the specific anatomical location and severity of brain pathology was often overlooked. Ultimately, the authors concluded that the relationship between MRI findings and motor outcomes needs to be further investigated in a cohort of children with CP using a valid, quantitative measure of MRI classification which includes detailed information about the location and extent of brain lesions, as well as valid and reliable motor measures [40,44].

The limitation of many cohort studies of children with CP in Canada [9], the USA, and across Europe [10] is the difficulty obtaining a representative sample and an entire cohort. The opportunity for undertaking entire prospective cohort based studies is possible in Australia. There is limited data on motor trajectories of an entire cohort of children with CP from diagnosis at 18 months to 36 months of age and these motor trajectories have not been correlated with MRI brain injury classification. For the present study the age of 18–24 months for entry has been chosen as diagnosis is usually confirmed by this time. Children will be followed up till 5 years of age at school entry when motor outcome has been well classified [3]. The preferred age for structural MR imaging is from 24 months because by this age myelination of the brain should be complete, thus allowing optimum differentiation between grey and white matter on MR imaging, important for the detection and correct classification of brain injuries and malformations (Figure 2).

In the Australian CP child study (NHMRC 465128) entire birth years of Victorian and Queensland born children with CP are prospectively entered and will be followed intensively to determine the relationship between the rate and limit of motor development (gross and fine motor function) as related to the nature of the brain lesion. Secondarily the influence of musculoskeletal deformity (hip displacement, spasticity and muscle contracture) and location and extent of brain injury will be related to the rate and pattern of motor disability. The parent report of their child's ability to participate in society and perceived quality of life will be compared across motor severity. Finally the level of motor functioning will be correlated with direct medical and allied health costs and outcomes including school readiness (see study flow chart, Figure 3). School readiness is a framework for assessing profiles of strengths and vulnerabilities of the preschool aged child [45]. It considers a child's readiness to learn within five major skill areas: health and physical development, emotional well-being and social competence, approaches to learning, communication skills, and cognitive skills and general knowledge [45].

Aims and hypotheses

This study aims to determine the pathway(s) to motor outcome (gross and fine motor) from diagnosis at 18 months to outcome at 5 years in relation to the nature of the brain lesion (using structural MRI). These aims will be explored through the following hypotheses:

1 The rate of motor development (gross motor function) from 18 months will be related to the limit of attainment at 5 years (Gross Motor Function Classification, GMFCS level).

2a The pattern of motor disability (motor type and distribution) will correlate with the location, presumed timing and nature of the brain lesion(s).

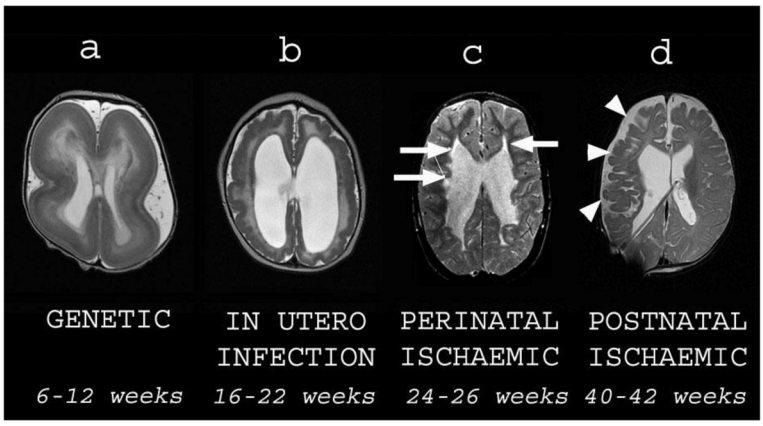
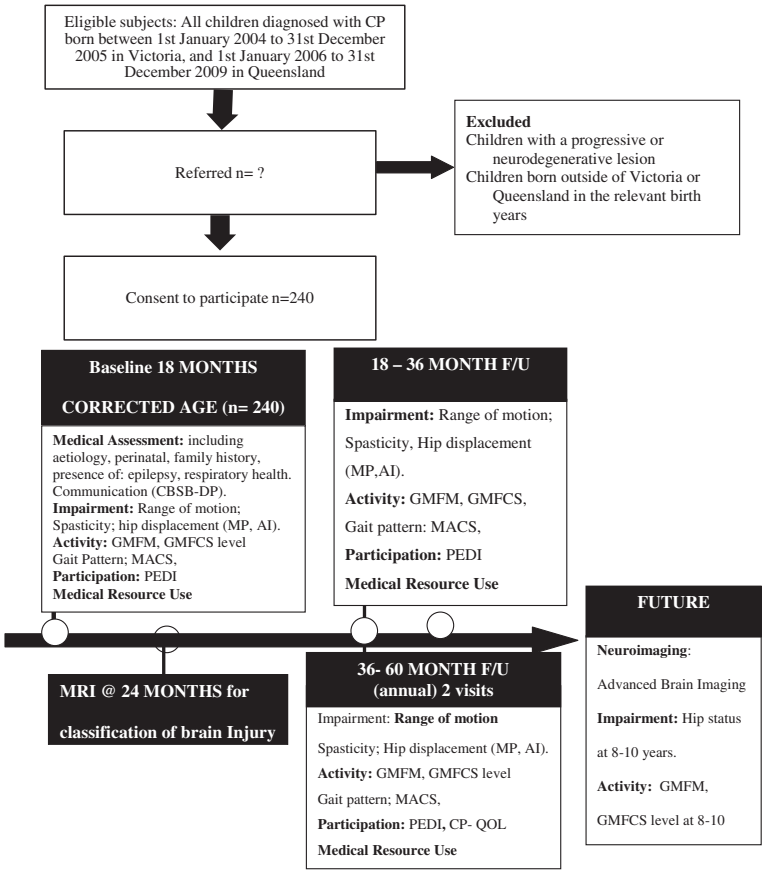


Figure 2 Examples of different types of structural brain abnormalities in cerebral palsy All images are axial T2-weighted MRI scans. Each image is subtitled by its presumed aetiology and timing during gestation. **a** is a child with lissencephaly showing cortical thickening and agyria. **b** is a child with congenital cytomegalovirus infection showing an overfolded cortex (polymicrogyria), thin white matter and dilated lateral ventricles. **c** is an ex premature child showing cystic white matter injury (arrows) consistent with periventricular leukomalacia. **d** is a child who suffered a haemorrhagic stroke in the newborn period. There is cortical and white matter loss in the right frontal and parietal lobes (arrowheads) consistent with previous ischaemia.



Legend: MRI = Magnetic Resonance Imaging; MP = Migration Percentage; AI = Acetabular Index; GMFM = Gross Motor Function Measure; GMFCS = Gross Motor Function Classification system; MACs = Manual Assessment Classification system; PEDI = Pediatric Evaluation of disability Inventory. CPQOL = Condition specific QOL measure (NHMRC 284514); Gait Pattern Classification.

Figure 3 Consort flowchart of study program.

2b The severity of motor disability in CP (age of onset or signs) will correlate with the location, extent and nature of the brain lesion (on structural MRI).
3 The rate and limit of motor development will be influenced by the severity of musculoskeletal deformity (i.e. slower motor development will correlate with marked hip displacement, increased spasticity and reduced range of motion in the lower limb).
4 Children with lower levels of function will have higher direct medical and allied health costs.

Study significance

This unique project will

1. Allow clinicians to better predict the functional outcomes of children with CP from an earlier age based on their rate and limit of gross motor abilities and nature and severity of their brain lesion.
2. Determine the nature and timing of physical deformities including hip displacement to guide the timing and intensity of interventions.
3. Provide comprehensive data on the relationship between the nature of the brain lesion, rate of musculoskeletal deformity and impact on the child's ability to participate in the community.
4. Information on resource use for future planning of medical and therapy services.

Methods

All children diagnosed with CP, born in the years 1st January, 2004 to 31st December, 2005 in Victoria, Australia and 1st January 2006 till 31st December, 2009 born in Queensland, Australia will be entered (n = 240). We define Cerebral Palsy as a permanent (but not unchanging) disorder of movement and posture that results from an insult to the developing central nervous system. The characteristic signs are spasticity, movement disorders, muscle weakness, ataxia and rigidity [43].

Exclusion criteria

1. Children with a progressive or neurodegenerative lesion.
2. Children born outside of Victoria or Queensland in the relevant birth years.

Ethics approvals

Ethics committee approvals have been gained through The Royal Children's Hospital Melbourne Ethics Committee, (HREC/25010 F), Southern Health Human Research Ethics Committee C (05077C), University of Queensland Medical Research Ethics Committee (2007001784), the Children's Health Services District Ethics Committee (HREC/07/QRCH/107), the Mater Health Services Human Research Ethics Committee (1186C), the Queensland Cerebral Palsy Register at the Cerebral Palsy League of Queensland (CPLQ 2008/ 09–1010), Gold Coast Health Service District Human Research Ethics Committee (HREC/08/QGC/45), Central Queensland Health Services District Human Research Ethics Committee (HREC/08/QCQ/19), Cairns and Hinterland Health Service District Human research Ethics Committee (HREC/08/QCHHS/521) and the Townsville Health Service District Human Research Ethics Committee (HREC/08/QTHS/33). There are no known health or safety risks associated with participation in any aspect of the described study. All families will give written informed consent to participate, and they are able to withdraw their child from the study at any time without explanation, without any penalty from staff at the Royal Children's Hospital or University of Queensland, or any effect on their child's care. Data collected in this study will be stored in a coded re-identifiable form (by ID number). Each child has multiple assessment appointments across the duration of the study, which necessitates data to be re-identifiable.

Ascertainment of the cohort

Prospective entry of birth years born in Victoria (born in 2004 and 2005) and Queensland (born in 2006, 2007, 2008, 2009) entered at 18 months will be followed until school age (5 years) (n = 240-360). Study recruitment commenced in July 2005 (at 18 months c.a.) for children born in January 2004 and continues in Queensland according the above birth years.

State wide recruitment has been established in collaboration with the relevant Cerebral Palsy Registers with data collection at tertiary referral hospitals. Community awareness has been generated through campaigns aimed at paediatricians (Division of Paediatrics & Child Health), general practitioners, allied health professionals, maternal and child health nurses, and neonatal follow-up clinics. These groups have been encouraged to refer children with motor delay (not sitting at 10 months, not standing at 12 months not walking at 24 months) for confirmation of a diagnosis of cerebral palsy. Families of children identified through the relevant CP Register have been approached after permission to contact the family has been given by their treating clinician or direct referral to the study by families whom have provided consent to be entered onto the Queensland CP Register (QCPR). Specialist clinics have been established at the tertiary referral centres where suitability for the study can be confirmed. In cases where the diagnosis of CP is unclear, or where there is a suggestion of a progressive or degenerative course, further investigations (such as metabolic screening) will be requested before a diagnosis of CP is confirmed. Parents have then been invited to participate

in the study and give informed consent. High ascertainment is expected for children with moderate to marked motor delay (GMFCS III to IV) and this has been the case for children born preterm and children referred to surveillance clinics at tertiary referral centres. Children born at term with mild motor delay (GMFCS level I, II) and predominant lower limb involvement (diplegia) are typically identified through the CP orthopaedic services and spasticity management clinics. Children with hemiplegia (GMFCS level I and II) are detected early through the surveillance clinics and occupational therapy services. Children who are detected after 18 months of age will be entered into the study at the time of diagnosis, will be offered brain MRI at entry and be followed up with serial motor assessments and other outcomes until outcome at 5 years.

Measurements and procedures

Following confirmation of a diagnosis of CP, eligible children are entered from 18 months corrected age. They will be assessed for diagnostic criteria, co-morbidities and for differential diagnosis by neurological assessment (by a Paediatrician, Child Neurologist or Paediatric Rehabilitation Specialist). Experienced Physiotherapy researchers will perform all GMFM assessments adjacent to either clinic visit and perform collection of range of motion, clinical measures of spasticity, then rate GMFCS, gait pattern, MACs and measures of pelvic radiographs according to standardized protocols.

Primary measures

The aim of the present study is to gather information regarding the longitudinal measurement of Gross Motor Function (GMFM-66) from 18 months to 5 years [46] and determine the aetiology of CP using clinical evaluation combined with MRI (location, nature and structure of the brain lesion) [27]. The lesion will be classified by 3 main criteria:

A. the *anatomical features* of the lesion:
   i. localisation by tissue (e.g. cortical, white matter, deep grey matter etc.)
   ii. localisation by region (e.g. lobes involved, laterality etc.)
   iii. extent of lesion (e.g. generalised, hemispheric, lobar etc.)
B. the presumed *aetiology* of the lesion: (i) genetic; (ii) ischemic; (iii) infective and (iv) other.
C. the presumed *timing* of the insult that caused the lesion:
   i. Prenatal by trimester or by stage of brain development;
   ii. Perinatal;
   iii. Postnatal.

All MRIs will be classified by a neurologist together with a neuroradiologist using a standardised method of image evaluation and classification. Following these evaluations, consensus will be reached regarding the above three criteria. We estimate that 70–80 percent of children currently receiving a diagnosis of CP will have had brain MRI as part of their clinical work-up. The American Academy of Neurology has concluded that a brain MRI should be part of the diagnosis of CP in a previous practice parameter [37]. For Victorian patients, the majority will have had their imaging performed and reported through the Royal Children's Hospital, Melbourne or Monash Children's Hospital Medical Imaging Department on a GE Signa Echo Speed 1.5T MR scanner. For Queensland patients, the majority will have had their imaging performed and reported through the Royal Children's Hospital, Brisbane Medical Imaging department on a GE Signa Echo Speed 1.5T MR scanner. The current minimum imaging protocol for patients with suspected CP consists of axial fast spin echo and coronal fast spin echo sequences and 3D inversion prepared fast spoiled GRASS sequence. 3D acquisitions are reformatted in axial, coronal and sagittal planes, with additional oblique and curved reformatting. Age specific protocols are used to maximize the ability to detect cortical and white matter abnormalities at different stages of myelination. All existing neuroimaging will be re-reviewed by a neurologist familiar with the features of lesions that result in CP, most commonly either white matter injury or congenital malformations. A protocol will be used to describe the features of each patient's abnormality. The patient's imaging will then be classified using a system, which takes into account anatomical features, aetiology and presumed timing of the "insult" causing the abnormalities. If no MR imaging has been performed, or if previous imaging was only CT scans or poor quality MRI scans, then an attempt will be made to perform high quality MR imaging. Such imaging will usually be necessary for clinical reasons to be able to make an accurate diagnosis and exclude causes of CP that may have genetic implications for other family members. This approach is consistent with recent guidelines suggesting that all patients with the label of CP have high quality MR imaging on at least one occasion [37]. For children scanned prospectively, this will be performed at the either Paediatric Magnetic Resonance Imaging Centres. All MRI scans will be performed clinically under anaesthesia after informed consent.

Brain lesion severity will be assessed using a structured scoring proforma [44] based on the CH2 template [47], a highly detailed single-subject T1 template in MNI space, which is the international standard for brain mapping (International Consortium of Brain Mapping - ICBM). Lesions will be transcribed onto the proforma and the following measures obtained: number of (i) anatomical

lobes involved, (ii) number of slices on the template that were affected and (iii) size and distribution of the lesion measured by a global lesion score and lesion subscores. The number of lobes and slices affected will be the average of summed right and left hemispheres. To calculate total lesion score, each frontal, parietal, temporal and occipital lobe will be first considered in three sections: periventricular, middle and subcortical matter. Each section will be scored as 0.5 if less than 50% of area was involved; or 1, for greater than 50% involvement, with a maximum lobar score of 3. Lobar scores for each hemisphere will be summed, with a maximum hemispherical score of 12 possible. The total lesion score will be the sum of right and left hemispherical scores (maximum score 24). A 1-point score (involved/not involved) will also be attributed to 16 anatomical structures including the corpus callosum, the cerebellum and the main subcortical structures. The final maximum score of the scale will therefore be 40 (24 + 16).

Gross motor function

At each assessment gross motor function is evaluated using the GMFM-66 & GMFM-88 [46]. The GMFM-88 assesses childrens' motor abilities in lying to rolling, sitting, crawling to kneeling, standing, walking, running and jumping. The GMFM-66 is comprised of a subset of the 88 items identified (through Rasch analysis) as contributing to the measure of gross motor function in children with cerebral palsy. The GMFM-66 will be used to provide an overall measure of gross motor function and the GMFM-88 domain scores to explore specific motor skills [46]. Measures of GMFM will be rated by experienced research physiotherapists.

Secondary measures

Gross motor function classification system (GMFCS)

The Gross Motor Function Classification System (GMFCS) is a five level classification system of children's functional gross motor severity. It is based on self-initiated movements, anti-gravity postures and motor skills expected in a typical five year old [25,26]. Children who are independently ambulant are classified as GMFCS I or II, those requiring an assistive mobility device to walk classified as GMFCS III and those in wheeled mobility as GMFCS IV and V. Two physiotherapists, trained in the use of the GMFCS, independently observe and classify children in one of five functional categories [25]. The GMFCS has internationally established validity, reliability and stability for the classification and prediction of motor function of children with CP aged 2–12 years [24,25]. It has a high inter-rater reliability (generalisability coefficient = 0.93) [25]. Classifications of gross motor abilities change with age, therefore separate descriptions are used for different age bands. In the current study, the <2 years and 2–4 year descriptions are used. Lower inter-rater reliability is documented for the <2 years age band ($\kappa = 0.55$), as younger children's gross motor abilities are more variable, and less developmental information is available on which to base the classification [48]. The intra-rater (test retest) reliability from <2-12 years appeared to be acceptable (generalisability coefficient = 0.68). The GMFCS has been correlated with a number of motor scales, as well as CP distribution and type of motor impairment [49].

Motor type & distribution

Motor type of CP will be classified as spastic, dystonic, ataxic, hypotonic, choreoathetosis, mixed CP or unclassifiable according to SCPE guidelines [28,50]. Distribution will classified by number of limbs impaired (hemiplegia, diplegia, triplegia, quadriplegia) by at least two independent raters [51].

Motor performance

Functional performance will be scored on the Functional Mobility Scale (FMS). This is a valid and reliable measure of a child's usual walking ability at three distances (5 m, 50 m and 500 m), representing their home, school and wider community [52].

Gait pattern classification

Gait patterns will be classified according to the Rodda & Graham's Classification for spastic diplegia [53,54], which has demonstrated validity and reliability [53]. From least to most severe these were: (i) True Equinus, (ii) Jump Knee, (iii) Apparent Equinus and (iv) Crouch Gait. For children with unilateral CP, gait patterns will be classified according to Winters & Gage [55]. This classification considers the sagittal plane joint movements. Group I: foot drop during swing phase (Apparent Equinus). Group II: persistent ankle dorsiflexion (True Equinus). Group III: maintained plantar flexion through gait cycle plus limited knee flexion-extension. Group IV: similar to III, plus reduced hip flexion-extension [53,56]. Winter's classification [55] has good inter-rater reliability using written reports (weighted kappa, $w\kappa = 0.76$) and videos ($w\kappa = 0.63$) [57,58].

Upper limb function

Upper limb function is classified using the Manual Ability Classification system (MACs) [59]. The MACs is an international system to classify hand function based on the child's typical performance when handling objects in daily activities. This classification system was developed for children aged from 4–18 years, but has been shown to have good reliability for use in children as young as two years [59].

Radiological measures of hip displacement

Hip surveillance, including anterior-posterior (AP) pelvis x-ray, is recommended for all Australian children with CP to facilitate early detection and treatment of severe or progressive hip displacement [14,60,61]. The migration percentage (MP) is widely accepted as the gold standard measure in hip surveillance [12,62], measuring femoral head subluxation. Other measures include the acetabular index (AI), assessing acetabular dysplasia [63], and the femoral neck-shaft angle (NSA) [64,65]. As the pelvis and its radiographic appearance changes between birth and skeletal maturity [66], early surveillance may be impacted by bony growth and ossification, particularly if measurements are based on landmarks that are difficult to identify or absent in the immature skeleton. The reliability of migration percentage has been investigated in relatively small studies to date [67,68], and reliability data in very young children is infrequent. Hilgenreiner's Epiphyseal Angle (HEA) [69] is a radiographic measure describing the proximal femoral epiphysis and has been previously applied to assessment of coxa valga [70,71], but may offer prognostic information for hips at risk in cerebral palsy. It is the acute angle between a line drawn parallel to and through the proximal femoral epiphysis and Hilgenreiner's line [69].

Musculoskeletal development

A comprehensive musculoskeletal examination will be performed by paediatric physiotherapists recording data relating to joint range of movement, muscle length, leg length difference, bony anomalies, motor type and muscle contracture.

Clinical history and examination

At study entry including a comprehensive clinical history and examination at study entry is performed by a paediatrician, child neurologist or rehabilitation physician. The following information is collected:

a. Presence or absence of vision impairment, hearing difficulties; epilepsy;
b. Feeding issues including presence or absence of gastrostomy tube and failure to thrive;
c. Respiratory difficulties including episodes of pneumonia and aspiration;
d. Speech and language development.

Participation

Children's participation will be assessed (i) via parent-report on the domains of self-care, mobility and social functioning using the scaled scores of the Paediatric Evaluation of Disability Inventory (PEDI) which has good validity and reliability [72-74] and (ii) parent perception of health related quality of life using a condition specific tool the CPQOL-child by parent report [75,76] at 5 years.

Medical and allied health resource use

In order to determine the relationship between motor prognosis and medical and allied health resource use, the direct costs of treatment will be monitored and compared to outcomes with adjustment for confounders such as disease severity.

Communication

Communication difficulties will be examined by parent self-report on the Communication and Symbolic Behaviour Scales–Developmental Profile (CSBS-DP) Infant-Toddler Checklist [77,78] (24 parent rated items) and the Communication Function Classification System (CFCS) [79]. The CSBS-DP screening tool is a parent questionnaire comprised of three composite subtests: social, speech and symbolic, and a total score. The social composite, composed of 13 questions, investigates the child's ability to functionally communicate, use eye gaze and gesture. The speech composite, comprising five questions, examines the sounds and words the child uses and their ability to combine words. The symbolic composite, comprising of six questions, explores the child's understanding of language and their ability to appropriately use objects such as a cup, spoon, toy telephone, stacking blocks, and participation in pretend play. Raw scores for each composite were converted into standardized scores (SS) where the M = 10 (standard deviation, $SD \pm 3$). The total score for the CSBS-DP was calculated by adding the raw composite scores, then converting to SS with M =100 ($SD \pm 15$) [77]. The CSBS-DP manual recommends all children with SS ≤ six on composites, or ≤ 81 on the total score, be referred for further speech and language evaluation. The CSBS-DP Infant-Toddler Checklist has been shown to have high test-retest reliability (r range = 0.79 to 0.88) [77], a strong predictive relationship with expressive and receptive language (R = 0.55 and 0.71 respectively) and high sensitivity and specificity (76% and 82% respectively) at two years of age [77,78]. The Communication Function Classification System (CFCS) will be used to classify everyday communication performance of individuals with cerebral palsy into five classification levels [79]. All methods of communication performance are used in assigning the level of function, including both informal (gesture, behaviour), and formal (speech and symbolic communication systems). The classification has good inter-rater reliability, conducted on 69 children aged 2-18 years (0.66 overall, and 0.77 for children older than 4 years), and excellent test-retest reliability (0.82) [79].

Neurological Examination: Existing data regarding the child's neurological examination will be reviewed.

Children will receive a comprehensive neurological examination by a rehabilitation specialist, developmental paediatrician or paediatric neurologist. It will be undertaken again if this has not been performed or documented comprehensively by such specialists within the previous six months.

Epilepsy

Epilepsy is common in CP, occurring in around 50% of children [80-82]. The presence of poorly controlled epilepsy or excessive anticonvulsant medications may confound an accurate assessment of each child's clinical state. For this reason we will obtain data on each child's pattern of epilepsy including age of onset, seizure type, frequency and medications.

Data analysis plan

A comprehensive database has been established for all data collection, including clinical measures, MRI scoring and questionnaires so that it is entered prospectively at the time of each assessment. Summary reports are automatically generated from the database to report back to families and treating clinicians after each visit. Our biostatistician will supervise the statistical methods proposed in this study, including analysis of binary outcomes in longitudinal studies using weighted estimating equations (e.g. presence of co morbidities); multilevel mixed-effects models of longitudinal binary outcomes (e.g. GMFCS levels), and generalised estimating equations for ordinal data.

For hypothesis I: Raw GMFM total score will be converted to GMFM-66, Rasch analysed scores. The GMFM-66 data will then be plotted by age in months for the entire cohort then according to GMFCS group. Parameters of a non-linear model of motor development will be estimated using non-linear fixed effects modelling for children according to their GMFCS level. The model uses two parameters, the estimated rate and limit of motor development. Other complex, longitudinal analysis methods such as multilevel mixed-effects models and generalised estimating equations [83] will also be employed to look at the temporal relationships between motor trajectories and classifications of brain structure on MRI (Hypothesis 1, 2), and musculoskeletal deformities (Hypothesis 3). For Hypothesis 4 groups of children (by GMFCS level) will be compared economically by incremental cost effectiveness and cost utility ratios.

Sample size calculations

For Hypothesis 1 six measurements are planned for each participant between 18 months and 5 years of age. A sample size of 40–50 per group (GMFCS I-V will give a total of 240 patients) for a two-group comparison of slopes in a linear model of motor development will have 80% power [9] of detecting if there is a difference between the GMFM curves based on initial GMFCS groups. This range allows for a range of possible effect sizes (based on results of Rosenbaum et al. [9]), and a range of between- and within-person variability in GMFM measurements over time (allowing for a linear pattern of motor development based on data from our own study of 90 children over 3 years (NHMRC 980753). The initial GMFCS classification is the primary predictor variable and GMFM-66 score at five subsequent time points will measure the pathway to motor outcomes. In the event that children are diagnosed after 18 months corrected age they will be entered at the age of diagnosis and will drop in to the study at entry. Previous ascertainment rates suggest that children will be identified by 2–3 years which would allow a minimum of 3–5 data points for analysis, appropriate for linear modelling.

For Hypothesis 2 for comparisons among MRI classification levels (anticipating 43% PVL brain loss, 16% BG damage, 16% cortical/subcortical, 12% malformation/miscellaneous, and 10% normal from [5], or comparisons among GMFCS levels (anticipating 36% level I, 16% II, 14% III, 16% IV, 18% V: [6]) we need a total cohort of approximately 250 children. For the non-linear model of motor development, sample size calculation is complex however 80 subjects per group with 4 GMFM measurements was sufficient to estimate the asymptotic limit parameter with precision ± 3 GMFM-points (width of 95% confidence interval) in a similar population [9]. A study of approximately 40 per group with 6 measurements will have slightly lower precision for this parameter but should be sufficient for identifying differences between GMFCS groups as the differences are large (>10 GMFM-points) [9].

Discussion

This study protocol describes the rationale, aims, hypotheses and methods for a large prospective longitudinal population-based study of early motor development and brain structure in a representative sample of preschool aged children with Cerebral Palsy, using direct clinical assessment. The results of this study will be published in peer reviewed journals and presented at relevant international conferences.

Competing interests

The authors declare that they have no competing interests.

Authors' contributions

RB is the chief investigator and together with MF and BR conceptualized, designed and established this research study. RB, RJ, AM, CF, BL, PB and MK also contributed to study design and were responsible for the selection of particular assessments. RB, MF, LM and AG were responsible for the brain MRI analysis content. RB, LP were responsible for ethics applications and reporting. RB, RJ, LP, LM, MK, MW will be responsible for recruitment and data collection in Queensland and RB, AM, MF, BR for recruitment and data

collection in Victoria. RB drafted the manuscript with input from all the co-authors. All authors have agreed the final version of the manuscript and were involved in the decision to submit the manuscript. There is no financial support for the authors regarding this manuscript. The external funding agencies (NHMRC, Telstra Foundation) have provided funds for the conduct of the study but will not be involved manuscript preparation, decisions to publish or the interpretation of results arising from the study. All authors read and approved the final manuscript.

Funding statement
Funding for this study has been received by the National Health and Medical Research Council of Australia for Project grant: 465128, NHMRC Career Development Fellowship, (RB) 1037220 and the Telstra Community Fund (Charitable Foundation).

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Received: 6 March 2013 Accepted: 31 May 2013
Published: 11 June 2013

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doi:10.1186/1471-2377-13-57
Cite this article as: Boyd *et al.*: Australian Cerebral Palsy Child Study: protocol of a prospective population based study of motor and brain development of preschool aged children with cerebral palsy. *BMC Neurology* 2013 **13**:57.